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Exploring Systemic Therapeutic Approaches for Individuals with Intellectual Disabilities using a Narrative Synthesis of Existing Research and Interpretive Phenomenological Analysis: A Portfolio Thesis

Lindsey Eunson

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Lay Summary of Thesis

Name of student:	Lindsey Eunson	UUN	S1061346
University email:	S1061346@sms.ed.ac.uk		
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This portfolio thesis is presented in two sections. The first includes a review of existing literature, that explores the outcomes of using systemic therapeutic approaches for individuals with intellectual disabilities and additional mental health or behavioural difficulties. Intellectual disability is characterised by significant deficits in intellectual functioning (IQ below 70), and adaptive functioning, which involves everyday social and practical skills. Historically, this set of deficits would be labelled mental retardation, or more recently, learning disability. People with intellectual disabilities are at a greater risk of developing several mental health and physical health problems, in comparison to the general population. This often leads to greater dependence on their support networks. As the government looks to reduce the number of people with intellectual disabilities being placed in long-stay hospital beds, more people are continuing to live with family

carers, often well into adulthood. This change places a huge amount of stress on those left with caregiving responsibilities, who are also then at greater risk of developing mental health and physical health difficulties associated with their role. Systemic approaches are being increasingly utilised with these families. Relationships are used as a resource in systemic approaches, to reduce stress, improve problem-solving abilities, and improve communication skills, in families or 'systems'. These systems may involve family members alone, but often involve other individuals such as support workers, mental health professionals, and teachers. The outcomes of relevant studies are described in this review, with careful consideration of the quality of each study, as well as their strengths, weaknesses, similarities, and differences. Recommendations for future research is provided.

The second section takes the form of a study, which explores the use of a specific type of systemic therapy called Behavioural Family Therapy (BFT), when used with individuals with intellectual disabilities and their families or support systems. More specifically, this study explores caregivers' experiences of being a BFT participant. Eight caregivers from three Scottish Health Boards; NHS Lanarkshire, NHS Lothian, and NHS Ayrshire and Arran, were recruited by their local Intellectual Disability Team Lead, to take part in a semi-structured interview. Candidates were selected because they had received BFT through the service that supported their relative with an intellectual disability and additional behavioural or mental health difficulties. Caregivers had all attended at least 5 sessions of BFT and had progressed beyond the engagement phase of the intervention. The interview involved open questions about their experiences of being a BFT participant, which allowed freedom to discuss anything that was significant to them. Interviews were analysed using Interpretive Phenomenological Analysis, which aims to uncover a rich understanding of the meaning that caregivers assigned to

their experiences. Interviews were transcribed, and read several times, to help the author to immerse themselves in the experiences of these caregivers. Similarities and differences between caregiver narratives were considered, which involved a deep level of interpretation. Significant themes emerged from this analysis, which were presented in relation to existing research; 'journey to acceptance and engagement in BFT', 'sense of progression and change', and 'caregiving in a challenging system'. Clinical implications and recommendations were discussed, and areas for future research were proposed.

Research Portfolio Abstract

Background: This thesis portfolio comprises two main sections. The first involves a systematic review exploring the outcomes of systemic approaches used for individuals with intellectual disabilities with additional mental health or behavioural needs, and their support systems. Intellectual disability in one member of a system can have an extensive impact on those around them, and often leads to greater dependence on support systems. It is therefore important that we understand how to adequately support the system and sustain this resource. This systematic review sought to explore and synthesise the outcomes of various systemic approaches used in this context.

The second section takes the form of an empirical study to explore a specific systemic intervention. The impact of intellectual disability often leaves their carers at greater risk of stress and mental health difficulties. Behavioural Family Therapy (BFT) is a form of psychoeducational family intervention that was developed to reduce stress and improve communication within families. This study aimed to explore the experiences of primary caregivers for an individual with intellectual disabilities and additional mental health or behaviour difficulties, related to being a BFT participant.

Method: The systematic review involved a thorough search of online databases and reference lists to identify relevant articles, as defined by predetermined eligibility criteria. 12 articles were identified, which were synthesised and evaluated using an adapted quality rating scale recommended by CASP and SIGN guidelines checklists.

The empirical study involved engaging eight primary caregivers in semi-structured interviews about their experiences of being a BFT participant. Interviews were transcribed and analysed using interpretive phenomenological analysis.

Results: The systematic review highlighted the potential benefit of using systemic approaches with individuals with intellectual disabilities and their support systems. Several articles reported on improvements in the behaviours of multiple family members, not just the individual with the intellectual disability. Many also noted improvements in the communication and interactions of system members as well as overall improvements in relationships. However, uncertainty regarding authors' definition of 'systemic approach' made it difficult to ascertain the degree of true systemic influence within evaluated interventions. Furthermore, the overall quality of the reviewed articles was poor.

Three superordinate themes emerged from the empirical study analysis; 'journey to acceptance and engagement in BFT', 'sense of

progression and change', and 'caregiving in a challenging system'. A fear of judgement permeated caregiver narratives, which made openness challenging. They felt that trust, empathy and validation were essential elements in fostering a sense of safety and comfort during disclosure. Caregivers also speculated on the timing of intervention, some feeling that crisis enabled engagement, whilst others felt crisis disabled their ability to attend to and engage in intervention. Caregivers all reflected on their sense of adjustment and benefit from BFT, with many reporting increased confidence in their caregiving abilities. Lastly, all caregivers referred to the current, consuming stress, and worry they were experiencing. This stress demanded vast amounts of their attention, making it difficult to engage in day-to-day tasks. This also led to difficulties in reflecting during interview. Several reported increased cynicisms about the world and the people in it, as a result of their experiences and their sense of duty to protect the individual with intellectual disability. These considerations were conceptualised in light of their expectation to engage in and facilitate change in BFT.

Discussion: The evident paucity of quality literature in this area indicates the need for more research to build upon the limited evidence base. Poor study quality often resulted from the study design, their use of outcome measures (not validated for the appropriate population) or lack thereof, and ambiguities relating to

defining systemic approaches. More methodologically sound research would help to resolve these issues and may aid a better understanding of the useful 'active ingredients' of systemic intervention for use with individuals with intellectual disabilities and their support systems.

The empirical study also highlights the benefit of further research into the use of BFT in this population, though the results do indicate that caregivers found elements of this intervention helpful. Results are discussed in relation to existing research and current policy. Limitations are acknowledged and clinical implications are discussed, which include the importance of developing therapeutic trust, the benefit of engaging the whole system as opposed to just two members, and the need to acknowledge the severity of stresses impacting the system's ability to accept and engage in BFT. Future research exploring the experiences of individuals with intellectual disabilities of being a BFT participant, would nicely complement this study. Larger quantitative research may help to determine whether objective changes, as postulated by caregivers in this study, may occur at group level.

Table of Contents

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Outcomes of systemic therapeutic approaches for individuals with intellectual disabilities: A systematic review

Running Title: Systemic therapies and intellectual disabilities: A systematic review

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Abstract

Background: The impact of intellectual disability spans wider than the individual alone. To account for this, an increasing amount of psychological interventions are embracing a systemic approach. However, research to support the efficacy of systemic therapies for this population is scarce.

Objectives: To synthesise and explore the quality of research to support the use of systemic therapies for individuals with an intellectual disability and additional behavioural or mental health needs.

Methods: 12 papers were selected from a thorough search of online databases and reference lists, according to specific eligibility criteria. Findings were synthesised and evaluated in line with pre-defined criterion for rating quality adapted from CASP and SIGN guidelines checklists. The review was prospectively registered on PROSPERO: CRD42018107673.

Results: 563 articles were extracted, 12 met the inclusion criteria; 1 RCT, 2 controlled trials, 7 uncontrolled studies, and 2 case studies. Outcomes indicate utility in employing systemic approaches for intellectual disabilities, though ambiguities around operational definitions of 'systemic' existed. The main benefits reported relate to

improved behaviours in all members of the system, improved communication, interactions, and relationships in general.

Conclusions: This review highlights the potential benefit in using systemic approaches with individuals with an intellectual disability. However, the overall quality of the articles is poor, largely due to study design and use of unvalidated outcome measures. The evident paucity of literature in this area indicates the need for more quality research to build upon the limited evidence base.

Introduction

Approximately 1% of the global population are diagnosed with an intellectual disability, although this rate is almost doubled in low- and middle-income countries compared to high income countries (Maulik et al, 2011). Diagnosis requires assessment and confirmation of deficits in both cognitive and adaptive functioning evident from childhood. Intellectual disability is aetiologically heterogeneous in nature and has both environmental (Patel et al, 2010) and genetic (Leonard & Wen, 2002; Chiurazzi et al, 2007; Ropers & Hamel, 2015) causes, though the origin is uncertain (Rauch et al, 2006) or a mix of the two (D'Amelio et al, 2005) in many cases.

Co-morbidity is common in people with intellectual disabilities, who are at greater risk of experiencing several mental health difficulties (Cooper et al, 2007) and physical conditions such as gastro-

oesophageal reflux disorder (Böhmer et al, 1999), arthritis, cancer, diabetes, epilepsy, other cerebral vascular diseases, and multi-morbidities (Emerson et al, 2016). People with intellectual disability are also more likely to be obese, have reduced lung functioning, and be under prescription of five or more medications (Emerson et al, 2016). Although much of the literature focuses on older adult populations with intellectual disabilities (McCarron et al, 2013; Hermans & Evenhuis, 2014), research also indicates that children with intellectual disabilities are more likely to have psychiatric disorders (Emerson & Hatton, 2007).

The impact of intellectual disability on an individual is vast, often leading to greater dependence on family, friends and healthcare professionals. Indeed, we do not exist in a vacuum, but in an intricate web of relationships within context. The impact on the wider system around the person with intellectual disability is therefore profound and complex. Parents of children with developmental disorders report more physical health problems, stress, and less social support than age and gender matched-controls (Cantwell, Muldoon & Gallagher, 2014). Carers of adolescents with intellectual disabilities also report higher levels of stress (Patton et al., 2018). Families supporting a child with intellectual disability are more likely to suffer from depression (Olsson & Hwang, 2001), and are more likely to be economically disadvantaged compared to those who support

children without intellectual disabilities (Emerson, 2003). Given the wide spread impact an intellectual disability can have not just on the individual themselves, but the entire system around them, the rationale behind implementing a systemic approach to intervention for people with intellectual disabilities is clear.

Systemic approaches have been considered and utilised historically, largely because of the theory that people with intellectual disabilities are immune to emotional difficulties and impervious to the benefits of individual therapeutic intervention (Freud, 1953; Bender, 1993). Based on this assumption, it would seem beneficial to engage the system around the individual with intellectual disability in therapeutic interventions. This also dissuades the pathologizing of individuals by placing more emphasis on interpersonal events and issues (Fredman, 2010). One could assume that the goal of systemic approaches is the same as other individual therapies such as Cognitive Behavioural Therapy, i.e. to effect change in the individual. However, systemic approaches intrinsically necessitates involvement from other members of the system, therefore it is reasonable to hypothesise that 'change' may be invoked in any or all engaged participants.

Although systemic approaches can take various forms and could be construed to mean any therapy that involves more than one party,

this is not the case. Some authors subsume all systemic and family treatments under the term 'systemic intervention' (Carr, 2014; Pinquart et al, 2016), which would encompass a large portion of literature such as couples' therapy, and parenting programmes that involve their children. Indeed, a vast amount of existing literature on intervention in intellectual disability explores the use of applied behavioural analysis (ABA), whereby family members are often recruited to implement and reinforce new behaviours (Hudson et al, 2003; Heitzman-Powell et al, 2014). However, the mere involvement of another individual is insufficient to label it a systemic approach. The focus in systemic approaches is arguably more complex and involves consideration of more interpersonal interactions. The difference is sometimes subtle and relate to the intervention's theoretical underpinnings. Purely behavioural interventions rooted in learning theory, such as classical or operant conditioning involving more than one individual, are not of particular interest here, because they do not place specific focus on relationships. However, both interventions often coexist in treatment packages with systemic components of intervention. Instead, a broad definition of systemic therapy, similar to that adopted in other reviews of the literature (e,g, Kaslow et al, 2012; Retzlaff et al, 2013), has been used, as defined by the UK council for Psychotherapy (UKCP, 2019) (Table 1). This broad definition encompasses a wider scope of relevant

literature. However, it also clouds the distinction between unique elements of different specific therapies and their contributions to outcomes. For example, many interventions involve small components of traditional systemic therapy alongside other therapeutic approaches such as cognitive behavioural therapy and often medication, complicating the discernment of the specific mechanisms involved in any observed change.

The terms systemic or family approaches and intervention are used interchangeably in this review. They do not denote the use of systemic therapy in its pure form or any one specific therapy, but to therapies with a systemic orientation, informed by systemic theory. This reflects the terms and context of each corresponding study.

Table 1: Definition of Systemic Therapy (UKCP, 2019)

"Systemic therapies is a generic term for therapy dealing with people in relationship to one another, the interactions of groups, and their patterns and dynamics. **Systemic therapy has its roots in family therapy, and family systems therapy, and approaches problems practically rather than analytically. It does not seek to determine cause, nor assign diagnosis, but rather identify the stagnant patterns of behaviour within the group or family and address the patterns directly. The**

role of the therapist in systemic therapies is to introduce creative nudges to support the changing of the system, and address current relationship patterns, rather than analyse causes such as subconscious impulses or childhood trauma."

Systemic interventions, including family therapy and behavioural family therapy are similar in their emphasis on the relationships between each person within a system. Despite the name 'family therapy', it is not restricted to family members, but may involve individuals and their health care staff or support workers, or children and foster carers. Difficulties, or 'symptoms' are decentralised from one member and considered as derivatives from the system itself, in the form of mechanisms that inform repeated patterns of communication. This transactional process determines how members relate to each other and how they manage stress and conflict, which can then in turn reinforce the preceding patterns of communication. Some research suggests that the relational and transactional dynamics in families with intellectually disabled members differ to those without a member with intellectual disability (Kaslow & Cooper, 1978) and that this often presents as an increased prevalence of unhelpful repeated transactional processes. Systemic therapy aims to identify processes and where necessary, to adapt

them by exploiting the strengths and resources of each of its members. Its content aims to help people to talk about difficult issues in ways that respect everyone's experiences, and to help raise awareness and understanding of each other's needs. Change is considered in terms of the interactions between members, as this is considered an important factor in psychological wellbeing.

The potential for widespread impact of systemic intervention on various systems and/or subsystems complicates the formation of operational definitions of effectiveness, making it difficult to evaluate. Indeed, 'efficacy' and 'effectiveness' are measured in various ways throughout the literature. However, given that this approach is utilised throughout intellectual disability services, it is important that there is an adequate evidence base to support its utility. This systematic review aims to synthesise the existing evidence base for systemic approaches for individuals with intellectual disabilities and additional behavioural or mental health needs, and to explore the quality of research to support this model of working. This aim is kept broad to allow exploration into various methodologies exploited to investigate the utility, applicability, validity and various measures of effectiveness of systemic intervention for this population. It is hoped that this may indicate gaps in the existing literature and highlight areas for future research. It may also help to identify the most beneficial treatment pathways

evaluated to date, in supporting the health and wellbeing of the individual with intellectual disability and the system around them.

Method

Identification of studies

This systematic review was conducted in accordance with guidelines produced by the Centre for Reviews and Dissemination (CRD, 2009 http://york.ac.uk/inst/crd/). A review protocol was developed prior to commencement, which defined the review question, inclusion and exclusion criteria, search strategy, quality evaluation measures, and plans for dissemination (Table 2). The 'PICOS' framework (Population, Intervention, Comparators, Outcomes and Study Design; CRD, 2009) was employed to guide key inclusion and exclusion criteria.

Table 2: Review protocol

Research Question	To explore the outcomes of systemic therapies for individuals with intellectual disabilities and additional mental health or behavioural difficulties.
Inclusion Criteria	Studies that evaluate systemic therapies for adults or children with intellectual disabilities (evaluation of any kind using validated and non-validated measures).

	Studies that contain original data, these can include single case or case report design, small N designs, or larger trials. Studies available in English.*
Exclusion Criteria	Studies involving adults or children with Autistic Spectrum Disorder without an intellectual disability.
Search Strategy	Multiple database search; additional manual search for missed articles
Quality Evaluation Measures	The Critical Appraisal Skills Programme's guidelines (CASP; Public Health resource Unit, 2017) and Scottish Intercollegiate Guidance Network (SIGN) Checklist were combined and amended to form a measure that would evaluate this heterogenous group of articles appropriately.
Dissemination	Written in adherence to the Journal of Mental Health Research in Intellectual Disabilities author instructions.

* This is predominantly due to lack of translation facilities.

The following electronic databases were employed to locate existing research; Embase (1980 to 2019 Week 14), PsychINFO (1806 to April Week 1 2019), Ovid MEDLINER(R) and Epub Ahead of Print, In-

Process & Other Non-Indexed Citations, Daily and Versions(R) (1946 to April 5, 2019). Prospero was used to check whether any similar systematic reviews were currently being conducted. The Edinburgh University archive and ASSIA were also consulted. Search terms were devised by exploring possible synonyms for intellectual disability and for systemic therapy. Initial searches were conducted in August 2018 and again in April 2019. One additional article (Beasley et al, 2018) was included following the latter search. Reference lists of included studies were examined to identify any articles that had not been identified within the electronic search. The search terms used in all databases can be seen in Table 3. Additional search terms were explored in the initial stages. The final terms were chosen as they were most appropriately inclusive.

Table 3: Database Search Terms

Summary of Search Terms	
Systemic word search terms	"famil* therap*" OR "system* therap*" OR "famil* intervention" OR "system* intervention" OR "famil* approach" OR "system* approach"
Intellectual Disability word search terms	"development* disorder" OR "intellectual disab*" OR "learning disab*" OR "mental handicap" OR

	"learning difficult*" OR "mental retard*"

Data extraction & Analysis

Data was extracted from articles that met the inclusion and exclusion criteria and was then entered into an Excel spreadsheet, summarising the following information: title; author, year, country; participant demographic information; inclusion/exclusion criteria; study design and setting; intervention details; outcome measure information; and results.

Quality Assessment Tool

The CASP guidelines for appraisal of qualitative research and Randomised Control Trials were used to critically appraise each study (Appendix 1). Because no stipulations were made relating to methodological approach, guidelines were amended where necessary to adequately represent and assess the full range of research designs utilised in the final article pool. The CASP appraisal tool covers considerations for study quality appropriate for most methodologies and was therefore most suitable for adaptation. Amendments were based on those used in previous systematic reviews (Camargo et al, 2014; Rao, Beidel & Murray, 2008; White, Keonig & Scahill, 2007) and on the Scottish Intercollegiate Guidance

Network (SIGN) methodology checklists for controlled trials (Appendix 2). This variety of assessment tools helped to ensure that all quality evaluation considerations were present to assess diverse methodologies. To allow for a more objective approach to quality rating, core elements of the appraisal tools were collated and weighted according to quality as detailed in table 3.

CASP guidelines and the SIGN checklist for appraisal of systematic reviews were also applied to this review, to ensure all essential elements were present. All studies were appraised by the first author and a sample were also appraised by a second, independent evaluator.

Because of methodological heterogeneity, a narrative synthesis of findings was deemed to be the most appropriate way to describe the results.

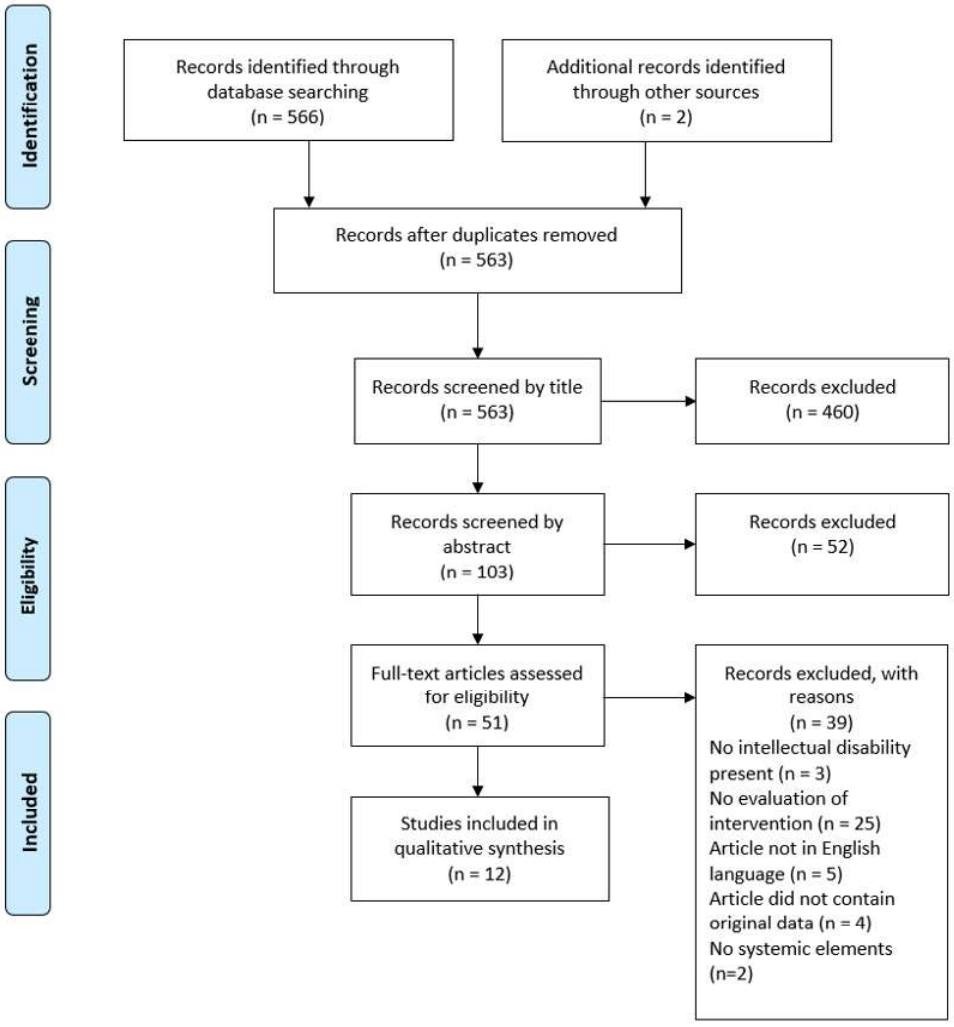
Results

Search Results

Results correspond to the April 2019 search. 566 articles were retrieved through the initial database search. ProQuest and ASSIA identified a further 2 studies. 5 were removed due to duplication. 460 studies were removed based on title alone; 205 were clearly unrelated to intellectual disability, 196 clearly did not involve systemic approaches, and 59 were reviews of the literature or did not

involve any evaluation or measure of outcomes. 52 were removed following screening of the abstract, 22 did not involve a systemic approach, 26 did not involve individuals with intellectual disabilities, 2 did not involve original data, and 1 involved consultation rather than direct intervention with the system itself. The full text of 51 articles were then assessed for eligibility. The search process is detailed in diagrammatic form in figure 1 below, based on the PRISMA statement (Moher, Liberati, Tetzlaff & Altman, 2009). The references for all included articles were checked for any additional relevant articles, though none were added.

Figure 1: PRISMA search process



Rating of study quality

The quality ratings for each study are summarised in Table 3 and the overall study quality are depicted in Graph 1 (see Appendix 3 for full scoring criteria). A Cohen's kappa of k=0.93 was obtained following independent evaluation of a small sample of articles. Following discussion, it was decided that an adaption relating to outcome measures was necessary to clarify the scoring criteria. For example, additional detail was added to the quality of outcome measures

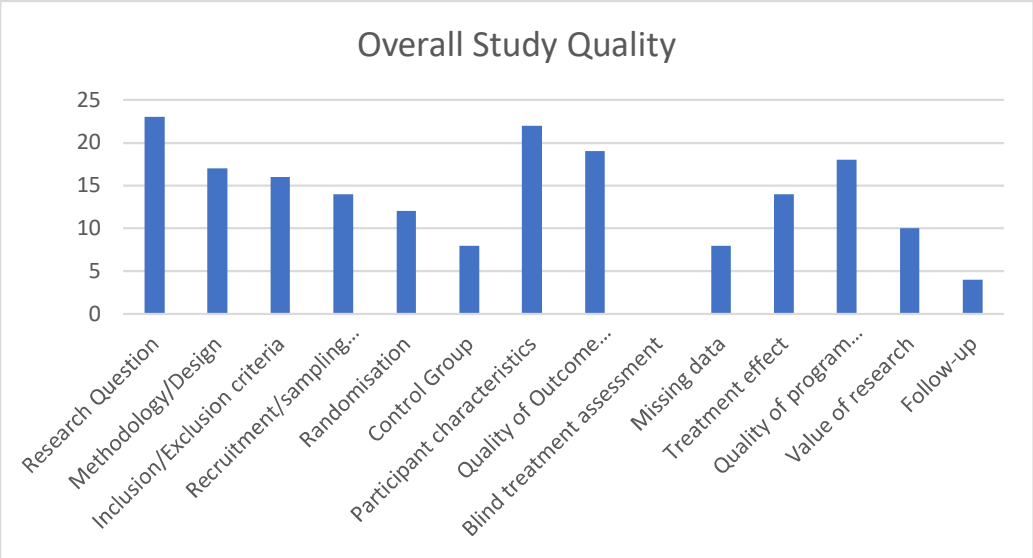
criteria so that the highest scores were given to studies that used outcome measures detailed with references, reliability and validity scores. This was in addition to previous criteria that stipulated that measures were validated, appropriate for use for people with intellectual disabilities, and appropriately addressed the study aims. The outcome measures section was then reassessed and 100% agreement on scoring was reached. Details of each quality assessment are summarised in Graph 1.

Table 3: Summary of overall study quality

Study	Design	Research Question	Methodology/Design	Inclusion/Exclusion criteria	Recruitment/sampling	Methods	Randomisation	Control Group	Participant characteristics	Quality of Outcome Measures	Blind treatment assessment	Missing data	Treatment effect	Quality of program facilitator	Value of research	Follow-up
Bagner & Eyberg, 2007	RCT	2	2	2	2	2	2	2	2	2	0	2	1	2	1	0
Hudson et al, 2003	CT Partially randomised	2	2	1	2	1	1	2	2	2	0	2	2	2	1	1
Kinast, 1987	CT Partially randomised	2	1	2	1	2	1	2	2	2	0	1	1	2	1	0
Beasley, Kalb & Klein, 2018	UCT, pre-post evaluation	2	1	1	1	0	0	2	2	1	0	2	1	2	1	0
Estreicher, 1982	UCT, pre-post evaluation	2	2	2	1	1	0	2	2	1	0	0	1	2	1	0
Philage, Kuna & Bercerril, 1	UCT, pre-post evaluation	1	0	1	1	1	0	1	1	0	0	0	1	0	0	0
Parker, Hill & Miller, 1987	UCT, pre-post evaluation	2	2	2	1	1	0	1	2	2	0	0	1	0	0	0
Lobato & Kao, 2005	UCT, pre-post evaluation	2	1	2	2	1	0	2	2	2	0	0	1	1	1	1
Glazemakers & Deboutte, 2	UCT, pre-post evaluation	2	2	1	2	1	0	2	2	2	0	1	1	2	1	0
Eisenhauer, 1991	Uncontrolled case series	2	2	2	1	1	0	2	2	2	0	0	1	2	1	1
Marshall & Ferris, 2012	Case Study	2	1	0	0	1	0	2	2	2	0	0	2	2	1	0
Mohamed & Mkabile, 2015	Case Study	2	1	0	0	1	0	2	1	1	0	0	1	1	1	1
		23	**17**	**16**	**14**	**12**	**6**	**22**	**19**	**0**	**8**	**14**	**18**	**10**	**4**	

CT (Controlled Trial)
UCT (Uncontrolled)

Graph 1: Summary of study quality assessment



Randomised Controlled Trials (RCTs)

RCTs are generally considered to be the 'gold standard' for intervention evaluation, though it is not always feasible to use this design. RCTs involve randomly allocating participants, or groups of participants, to an intervention group or a control group. Random assignment means you can more accurately estimate whether observable change is a consequence of the intervention, or other factors. The main challenges for RCTs is that they require large sample sizes, low attrition rates, and a largely homogenous cohort of participants.

Preliminary evidence for an unmodified parent-child interaction therapy used with children with intellectual disabilities was reported in the only RCT included in this review (Bagner & Eyberg, 2007). This

manualised intervention involves 60-minute, weekly sessions over approximately 12 weeks. The intervention has 2 phases, one of which focuses on enhancing the parent-child relationship, improving child social skills and increasing positive parenting. The other phase has a more behavioural focus, relating to skill development to improve boundary setting, and to maintain a consistent response to problem behaviours. Outcomes indicate that mothers in the experimental group interacted more positively with their child following intervention and that their interactions were more child-led, than those in the waitlist control group. Results suggest that mothers in the experimental group also used more positive behaviours and less negative behaviours than the control group post intervention. Behaviour change in mothers in the intervention group was statistically significant between time points, with effect sizes of d=2.06 for positive behaviours and d=1.32 for negative behaviours. Children in the experimental group were reported to be more compliant and exhibited fewer disruptive behaviours and behaviour problems than waitlist controls. Furthermore, a large effect size (d=1.53) was presented between Time 1 and 2 in relation to child compliance. Following further analysis, the authors concluded that increases in positive parenting interactions and decreases in negative parenting interactions mediated these behavioural changes. Lower parental stress was also reported in the experimental group

compared to controls, though this was not a significant change. Interestingly, maternal stress was relatively low at pre-treatment compared mothers of children without intellectual disabilities. Despite authors' conclusions, clinically significant, between-group change was evident on just two of the measures collected, however subsets of other measures and within-group comparisons did yield significant results. The measures are also limited by design in their ability to capture qualitative changes in parent-child interactions, which would offer further insight into overall systemic changes that may have occurred. Finally, the lack of follow-up limits the rigour of this study in terms of the sustainability of results.

Controlled Trials

Controlled trials involve the comparison of outcomes for two groups of individuals, ideally with similar baseline characteristics (matched controls), who have received different interventions. Prospectively, well-matched control groups can provide substantial evidence to support conclusions about the impact of intervention, though it is still possible to draw inaccurate conclusions.

Moderate effect sizes were observed in Hudson et al's (2003) controlled study exploring methods of delivery of their 'Signposts for Building Better Behaviour' programme. Their study examined the

utility of delivering this manualised, yet flexible intervention, which consists of a series of informative booklets with video supplements, in three different formats; via group, telephone, or self-directed. The authors recognised the importance of a systemic approach to traditional parenting training programmes that incorporates consideration of wider family needs. Broadly based on functional assessment of behaviour, Hudson and colleagues introduce additional stress management and problem-solving elements into their intervention that involve the whole family and the relationships within it. Participants were matched in terms of maternal education and occupation as well as child gender, age, and intelligence. No differences were observed between the pre-test scores of those who completed the intervention compared to those who did not. Outcome measures considered levels of depression, anxiety, and stress; sense of parenting competence; and perceived parental hassle and need. No differences were observed between the three different experimental modes, but improvements were identified across all three groups when compared to a waitlist control. Significant change across the combined experimental groups between pre-test and follow-up testing was identified in measures of disruptive behaviour (P=0.013) and antisocial behaviour (P=0.028). Effect sizes were not reported. Outcome measure and intervention completion was low, making it difficult to draw conclusions from this data alone. Although

careful consideration was made regarding methodology, randomisation could not be fully implemented due to concerns raised by participants allocated to the group condition relating to time commitments, logistical and geographical complications. These participants were subsequently reallocated to other groups to maintain study power. Although this study provides some evidence to support the utility of such interventions, this issue does weaken the quality of the research.

The second and final controlled study involves exploration into effective treatments for stress of parents of children with intellectual disabilities. Kinast (1987) investigated the effectiveness of parent-child play therapy and mutual group therapy in comparison to a control group. Effectiveness was measured by change in stress levels, conflict resolution skills, positive parenting behaviours, parents' perception of their child's maturity and skills, parental feelings of satisfaction and attachment to their child, and in parent/child relationships. The focus of interest for the purpose of this review was in relation to parent-child play therapy which involved modelling, instructions, practice and feedback of desired behaviours, education around behaviour management, play and child development, re-direction, and parent support. Again, a large proportion of the intervention is behaviour-based, however the author highlights the importance of enhancing the interaction and relationship between

the child and parent, whilst acknowledging the psychological needs and status of both individuals. Consideration of unhelpful parenting strategies and transactional dynamics were considered as part of the play therapy formulations, structure and content. Although the authors of this study imply a potential benefit of child-play therapy for parents of children with intellectual disability, the results yielded no significant results across all outcomes. A trend towards improved conflict resolution was however indicated. Attrition levels, the relative short duration of treatment, and the small sample of eight participants per group may account for this, although this is purely speculative. Further research is necessary to fully explore this possibility.

Uncontrolled case series; pre-post evaluation studies

Uncontrolled case series and pre-post evaluation can indicate whether significant change has occurred on any given measure, over a designated period of time. If participants are receiving treatment during this period, it may be predicted that treatment has contributed to any observed change, to some degree. The main problem for studies of this kind, are that without a control group, it is not possible to determine whether this change would have occurred over time without treatment. Although erroneous conclusions are

drawn at times, these studies can help to generate hypotheses about treatment effect, thereby highlighting potential areas for more robust research.

The most recent uncontrolled study included in this review was conducted in the USA by Beasley, Kalb & Klein (2018). They were investigating the use of a systemic intervention programme designed for people with intellectual disabilities and their wider, supporting networks (Systemic, Therapeutic, Assessment, Resources and Treatment; START). A sample of 41 individuals with intellectual disabilities and additional mental health or behavioural difficulties were recruited following engagement in the programme. This person-centred, strengths-based programme aims to enhance expertise and partnerships in the systems supporting individuals with intellectual disabilities, to improve overall wellbeing and quality of life. Authors aimed to determine changes in mental health symptoms following START, yet just one validated outcome measure was used (The Aberrant Behaviour Checklist; ABC). Furthermore, only three subscales were considered in their analysis; irritability, lethargy, and hyperactivity, and no outcomes were collected from parents or other health professionals. Psychiatric admissions were also considered in their results, though admissions prior to treatment were obtained via self-report. Their results revealed a significant reduction in all subscales of the ABC between pre and post measurements, with

moderate effect sizes for hyperactivity (d=0.58) irritability (d=0.62) and lethargy subscales (d=0.56). The proportion of individuals who experienced psychiatric hospitalisation, or who were seen in the psychiatric emergency department also reduced significantly between pre and post time points ($p<0.5$). Missing data was acknowledged to have a bearing on the obtained results and no follow-up was conducted to determine whether change was sustained. The START programme involves a variety of strategies to support families including crisis response, training and consultation. It is therefore difficult to draw concise conclusions about the systemic elements of this intervention in relation to changes in mental health.

Estreicher's (1982) uncontrolled study explored the use of family therapy for families including a 'developmentally handicapped' child. The aims and outcomes of the intervention were similar to those used in more traditional parent training interventions, but more focus was placed on the functional patterns of parent-child interactions and the level of involvement of parents with the identified child. For example, unhelpful patterns of interaction and behaviour may be identified, and adaptations may be gently encouraged with an emphasis on the strengths and positive qualities of each member of the system. Their results indicate that following family therapy, parents became more involved with the identified

child. Parents and siblings were perceived to communicate more, engage in more eye contact, and engage in more activities that involved the identified child. Their outcomes suggest that parenting style also changed following intervention, with more attentive, explanatory, demonstrative, and positive techniques being utilised. All members of the family reported to have found the intervention helpful and described feeling they had benefitted from it. These results provide preliminary support for the utility of family therapy for families that include a child with an intellectual disability, though no effect sizes or statistical changes were reported. Furthermore, this was an unpublished preliminary investigation that was not peer reviewed, and no follow-up was conducted to determine outcome longevity.

Philage, Kuna & Becerril (1975) report on a systemic treatment for children with intellectual disabilities, which also involves their parents and teachers in their uncontrolled study. Remediation and behaviour modification techniques were utilised with the aim of developing the relating skills of all members of the system and increasing independent functioning of the child. Individual goals were identified from each member as defined by their own perceived problem. Personal and interpersonal dynamics were addressed throughout to encourage greater understanding and empathy of each role within the system. Descriptive statistics of data collected

over 3 years provides some support for this intervention in a random sample of 4 subjective parental reports of behaviour and adjustment, suggesting improvement following intervention. Although the duration of this study is longer than all the other included studies, no group means were reported to indicate significant group changes over this period. Furthermore, standardized, parental reports are vulnerable to many biases. There was just one re-referral out of the 21 families involved. However, no indication of re-referral rates was provided for context, and no clear time frame was reported. Authors state that direct feedback was positive, but no detail about what form this feedback took was offered. Authors also report that their novel approach “seemed effective in changing behaviours” and that “each child changed positively”, but no discussion into what elements were responsible for evoking change or what the change looked like was reported. Authors did consider therapist impact, although no ethical consideration was documented. This is particularly interesting as part of the intervention involved children being removed from their families for 2 weeks with no personal contact permitted. Overall, this study indicates potential benefits of intervention, but limitations including the lack of information provided precludes any definitive conclusions being made. Emphasis on several systemic features was evident e.g. consideration of the relationships between all members, their thoughts and feelings

about each other and the process of intervention, how to share positive and negative feelings with each other, and exploration into how remediation may be meaningful to all parts of the system, as well as the whole.

Parker, Hill & Miller's (1987) results indicate a benefit in sharing difficulties with similar families. Their workshops were designed to teach communication skills and to foster consideration of wider issues within the system, that may contribute to overall stress or dysfunction in the family, i.e. not just difficulties identified within the child with intellectual disability. Their measures reveal mixed results in relation to self-esteem obtained via the Family Relationship Inventory (FRI; Michaelson & Bascom, 1978; Nash, Morrison & Taylor, 1982). Children with an intellectual disability viewed themselves more positively following the 6-month intervention period, however female siblings' self-esteem scores fell. Authors argue that this reflects a change to a more realistic view of the self. The FRI also calculates a total index of how individual family members are viewed by the family as a whole. These results were also mixed. Mothers' and sisters' view of the child with an intellectual disability was slightly more positive post treatment, while brothers' and fathers' view moved in the opposite direction. No ethical consideration was documented, despite potential distress arising from highlighting negative views of family members. Outcome

measure changes were not analysed statistically so significance levels were not reported, although this is likely due to the small sample size. Therefore, no firm conclusions can be made about the efficacy of this intervention for families of people with intellectual disabilities.

Siblings of children with intellectual disabilities were also considered in Lobato & Kao's study (2005). They reported on a group, family-based intervention called 'SibLink' for young siblings of children with chronic illness and developmental disorders. Six sessions of collateral, integrated sibling-parent groups were attended by participating dyads. Knowledge of their siblings' condition, their sense of connectedness with others in similar situations, and level of global functioning were explored. The first two sessions involved education and discussion to improve knowledge and understanding of their siblings' condition. The next sessions focussed on identifying and managing emotions, problem solving challenging situations, and identifying strengths in their sibling as well as balancing their siblings' needs within the needs of the family. Significant improvements were documented in siblings' knowledge of their brother or sisters' condition following intervention ($p<0.1$). Sense of connectedness also significantly improved ($p<0.1$), as did their perceptions of their own competence ($p<0.1$). No other significant results were reported.

A more recent relevant uncontrolled case series study (Glazemakers & Deboutte, 2013) evaluates a modification of the 'Positive Parenting Program' for parents with intellectual disabilities, delivered in group format. This intervention comprises one level of a multilevel system of behavioural family intervention (see Sanders, 2008). Many of the sessions are skills-focused using behavioural teaching strategies which are less systemic in nature. Yet, attention is also given to developing positive relationships and problem solving, and many sessions designate time for current issues, allowing opportunities to discuss relational difficulties. Reflection is encouraged and shared where appropriate throughout the intervention. Their results indicate reduced anxiety and general psychological stress ($p=0.043$) following intervention. Maladaptive parenting, and parent-reported child conduct problems also reduced post intervention with effect sizes of $d=0.62$ and $d=0.49$, respectively. Interestingly, parental report of child problems and their impacts worsened significantly following intervention ($d=-0.50$), though there is no mention of this in the narrative. Retention levels and satisfaction ratings were high in this study, which provides additional support for the authors' positive conclusions about this intervention. The main limitations of this study are its lack of follow-up, the inability to differentiate specific effective treatment components from confounding factors such as general

maturation, its limited recruitment methods, and biases that may arise from within service recruitment and self-report.

Eisenhauer (1991) explored change in families with intellectually disabled children following family systems therapy. The communication patterns of the family as a whole, and of the spousal subsystem were investigated, as were parental perceptions of their child's behaviour, classroom behaviour, and academic achievement. Ten, 60-minute, weekly sessions were delivered, initiated by a structural discussion tasks completely first by the parents alone, then by the family. Three families with a child with an intellectual disability and a control group of children in the same class as the identified child, formed the subject pool. A multiple baseline across-subjects design was used to explore intervention effects. This means that participants were randomly assigned to a three, five, or seven-week observation period to determine baseline characteristics. This design was used as withdrawal or reversal of existing treatment variables was deemed unethical or impractical. The results were mixed; small improvements were noted in academic performance and parent's perception of behaviours, which were of insufficient magnitude to reach statistical significance on a group level. Teacher's perception of child's achievement-compatible behaviours declined slightly. However, minimal but significant changes were reported in relation to family relational dynamics across various relational factors

following intervention. Authors concluded that communication patterns and relational dynamics in the parent dyad became more symmetrical (or equal) and that their interactions were more complimentary. Their interactive style also became more neutral, indicative of increased conflict-avoidance behaviours. Exploration into family relationships more generally reveals similar change, but with some increased or maintained competitive symmetry in terms of their interactions. The author provides elaborate detail regarding methodology, rendering it easily replicable, and great effort was taken to optimise the study's validity. The results of the study involve huge levels of analysis across different factors, therefore the study may be prone to Type II error. The results of this study are inconclusive in terms of intervention efficacy and the small sample size makes it difficult to generalise or to draw conclusions about specific intervention-related change.

Case Studies

Case studies can provide a rich, detailed account of an individual's experience of intervention. Individual perspectives and outcomes can provide valuable information that may indicate useful areas for further research. They may also help to form, refute or support hypotheses about an intervention. Case studies are particularly

useful when investigating rare occurrences, or when exploring new areas of research. The downsides to a case study design is that they often relies heavily on the researcher's subjective interpretations, the case example may not be representative of a wider population, and it is not possible to determine cause and effect.

Marshall & Ferris's (2012) case study is the first report known to date to explore the utility of Behavioural Family Therapy (BFT) for people with intellectual disabilities and additional mental health or behaviour problems. Their case example is a 45-year-old man with an intellectual disability, schizophrenia and autism. This intervention aimed to improve understanding of mental health issues through discussion and sharing of information. The service user was encouraged to lead discussions about how their illness affects them. The whole system was encouraged to set personal goals, and to seek assistance and support from other members to make these goals more achievable. This intervention focused on existing strengths of members of the system, to build on them, enhance problem solving abilities and promote positive communication. Results from a validated outcome measure indicate a reduction in caregiver strain and an improvement in family functioning following intervention. Again, as is the nature of case studies, this cannot be generalised, but provides impetus and rationale for further investigation into the efficacy of this approach. Minor amendments were made to standard

BFT delivery; therefore caution must be used when interpreting the results. Furthermore, BFT is known to invoke similar improvements in families that include a member with schizophrenia alone (e.g. Pharoah et al, 2006), therefore the positive changes could be attributed to improvements solely related to that diagnosis.

The final case study evaluates a structured, attachment focused, parent-child intervention to address relational difficulties thought to result in biting behaviours in a child with intellectual disability (Mohammed & Mkabile, 2015). Their assessment and intervention were based on the principles and techniques of 'Theraplay' child and family therapy (see Jernberg, 1984), which involves structure, engagement, nurture and challenge elements. Assessment identified a poverty in engagement, attunement, enjoyment, and mutual awareness of the other, therefore these areas became a focus for intervention. Reflective functioning and mentalisation skills were addressed and developed, which were considered catalysts for behaviour change in both parties. Their results are based on observations and reflections documented before and after intervention took place. This was an unplanned, retrospective research study, therefore no effort was made to control for extraneous variables that may contribute to observed outcomes. Observations indicate improvements in the mother's mentalisation skills, the overall quality of parent-child interactions, and a reduction

in child biting behaviours. These observations are reflected in parent self-report measures, however no objective measures were utilised, so it is not possible to confirm these claims, nor can one generalise the results. The study does however support the rationale for further investigation into the efficacy for such approaches for people with intellectual disabilities.

Summary of studies

This review explores the use of systemic interventions for people with intellectual disabilities and additional difficulties. An overview of the included studies is presented in Table 4. The most commonly reported difficulties presented, related to behavioural problems or challenges. Other additional problems involved dysfunctional parenting strategies, emotional or social difficulties, difficulties with maturational transitions, poor coping skills, stress and other mental health difficulties, negative attitudes, and attachment-related difficulties. Interestingly, one of the studies did not report on any additional needs of the person with the intellectual disability, indicating that this alone required specific intervention. Similarly, one study involved children with an intellectual disability and chronic illness, neither of which may be 'fixed' with treatment. The level of systemic focus varied across studies. Three of the studies adopted a purely systemic approach to intervention, while three involved a large systemic focus with behavioural and attachment-focussed

strategies included. Half of the described studies were structured largely around behaviour adaptation, but included additional elements of systemic intervention such as enhancing problem solving and communication skills using a strengths-based approach. A range of intellectual disability severity was explored in this selection of studies, ranging from mild to profound. Several comorbidities were also included, which is useful given the prevalence of comorbidity and multi-morbidity in people with intellectual disability, though may impact on results.

Table 4: Overview of included studies

Title	Study	Demographics	Inclusion/Exclusion	Study Design and setting	Intervention	Measures	Results
Parent-Child Interaction Therapy for Disruptive Behaviour in Children with Mental Retardation: A randomized control trial	Bagner & Eyberg, 2007 (USA)	30 female primary caregivers and their 3-6 year olds (77% boys). Mean age = 54.13 months. 60% had an IQ of between 55 and 75.All cohabiting family members were invited to participate but only 6 fathers accepted.	Severe to moderate mental retarcation and comorbic oppositional defiant disorder. Mothers must score over 75 on cognitive measure; ASD and/or sensory impairments were excluded as were chilcren suspected of having experienced abuse.	RCT with a waitlist control group. Research conducted in clinic, participant homes and classrooms	Manualised Parent-Child Interaction Therapy delivered weekly over an average of 12 weeks, lasting an hour each. Taught skills: enhancing parent-child relationships, increasing positive parenting, improving child social skills, setting limits, maintaining consistency.	Child Behaviour Checklist for 1.5 to 5 year olds (Achenbach & Rescorla, 2000); Eyberg Chilc Behaviour Inventory (Eyberg & Pincus, 1999); Parenting Stress Index: short form (Abidin, 1995); Dyadic Parent-Child Interaction Coding System (Eyberg, Nelson, Duke & Boggs, 2004); Therapy Attitude Inventory (Eyberg, 1993).	70% of the intervention group reached clinically significant improvement levels on the Child Benavicur Checklist, compared to 17% of the control group. 50% of the experimental group improved significantly on the Child Behaviour Inventory following intervention compared to 8% of the control group. Consumer satisfaction was high. A 47% attrition rate was reported. No follow-up recorded.
Evaluation of an intervention system for parents of children with intellectual disability and challenging behaviour	Hudson et al, 2003 (Australia)	115 mother-child dyads (34 female, 81 male) Age range: 4.6 to 19.4 years, mean 10.11 years. Children assessed as having an ID, IQ available for 72. 47% mildly disables, 49% moderately disabled, 4% severely disabled. 16% also had autism, 19% Down's Syndrome, 10% epilepsy, 11% visual impairment, and 10% cerebral palsy	Children with intellectual disability	Controlled Trial. Waitlist comparators formed the control group. Research conducted in participant homes and in clinic	Manualised signposting (group support, telephone support or self-directed). 6 fortnightly 120 minute group sessions in group mode facilitated by a therapist. Telephone mode: fornightly calls lasting 20 minutes on average. Participants were initially mailed the first two booklets, adjunctive booklets ard workbooks, the rest were delivered fortnightly. Self-directed mode: parents worked through materials with no support, same delivery method as the telephone group. Materials for all groups: 8 bcoklets, a videotape and a workbook. Skills taugh:: behaviour modification, teaching new skills, behaviour monitoring, dealing with stress.	The Parenting Sense of Competence Scale (Johnson & Mash, 1989); The Depression Anxiety Stress Scale (Lovibond & Lovibond, 1995); The Parenting Hassles Scale (Gavidia-Payne et al, 1997); The Developmental Behaviour Checklist (Einfeld & Tonge, 1989); The Consumer Satisfaction Scale.	57% overall completed outcomes. 28% of the experimental groups completed all outcomes including follow-up. No differences were identified between completers and dropouts. Significant improvements (moderate effect size) reported on all measures except the parental hassles scale and child behaviour subscale. No difference in effect size was identified between differert modes. Folllow-up 4-6 months post intervention, no follow-up for waitlist control as they were offered intervention following study completion.
A systematic investigation of effective treatment methods for the parental stress as related to the mentally retarded	Kinast, 1987 (USA)	11 males and 5 females aged 5 months to 60 months, average age pre-test = 3 years and 7 months. 5 males, 3 females in treatment group. Mild to profound deficits. 3 African American, 13 Caucasian. Mothers: 8 married, 2 single, 1 widow, 1 common law, 4 divorced.	Children needed some response to environmental stimulation, have basic sensory-motor schemas, and be able to be physically handles. Parents must be able to attend the facility.	Controlled trial. Control group contained 8 participants, who were assigned to the initial phase of the intervention. Treatment took place in a playroom facility.	12 Parent-Child Play Treatment sessions lasting 20-30 minutes were held weekly. Treatment consisted of modelling, instructions, practice and feedback of desired behaviours, education, play and child development, re-direction and parent support. Focus is placed on the child and the interactions in the dyad.	Conflict Tactic Scale; Parenting Stress Index; Adaptive Behaviour Scale for Infants and Early Childhood; Questionnaire of Expectations of the Child; Michigan Screening Parenting Profile	No significant relationship between level of stress and conflict resolutions. No significant treatment effect of conflict resolution between groups and between trials, but there was for all subjects between trials. No significant difference in overall stress levels between groups or in parents' expectations of the child's skills overall. No significant changes in parents' feelings of satisfaction and attachment to the child. No significant difference or changes in parents' relationships with others was found within or between groups. No follow-up.

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A new family approach to therapy for the learning disabled child	Philage, Kuna & Bercerril, 1975 (USA)	21 children, their parents and their teachers. 14 aged 6-9, 7 aged 10-13	Parents and teachers of children aged 6-13 with IDs, evaluative-contractual interview conducted to determine candidate suitability but no details documented	Uncontrolled case series, pre-post evaluation study. Intervention delivered in clinic, participant home, and classroom	4 non-manualised intervention programs; children with IDs aged 6-9, children with IDs aged 10-13, parent training, teacher training. Skills taught: behaviour modification; perceptual dysfunction of vision, audition, speech, coordination, hyperkinesis and resulting behaviour with a focus on remediation and socialisation. Relating skills were developed, i.e. synchronising feelings with words. Individual goals from each member were defined based on their own specific problem. Personal and interpersonal relaticnships were explored. Intervention consisted of 120 minute weekly sessions over 34 weeks.	Standardised behaviour and adjustment checklist; individual problem hierarchies; subjective information sheet for parent and child description of problems providing a problem severity quotient, standardised on a Likert scale. Questionnaires completed monthly by teachers.	76% of children completed all personal hierarchy problems, 95% children completed at least one. All four of the sample families reported reduction in problem severity quotients for both child and parent. 3 out of the 4 families reported a reduction in behaviour and adjustment difficulties, the other reported an increase on this measure. No follow-up was conducted.
Multiple family therapy: evaluating a group experience for mentally retarded adolescents and their families	Parker, Hill & Miller, 1987 (USA)	3 families; 16 years and 18 months, 9 year old brother, 9 year old sister, 39 year old mother and father; 20 year old adolescent, 12 year old sister, 40 year old mother, 44 year old father; 22 years and 2 months, 46 year old stepmother, 47 year old father.	a) family with adolescent aged 17-25; b) adolescent identified as 'educable mentally handicapped'; c) attended with an adult. Families excluded if chaotic they included an actively psychotic member	Uncontrolled case series, pre-post evaluation study. Intervention completed in clinical setting.	Non-manualised multiple family therapy group treatment. 90 minute sessions fortnightly for 6 months. Taught skills: communication, emotional problem solving, family functioning.	Family Relationship Inventory (Michaelson & Bascom, 1978; Nash, Morrison & Taylor, 1982);	Mixed results, child with ID viewed more positively following intervention and their self-esteem improved. Siblings self-esteem decreased following intervention and they were viewed less positively by other family members. No follow-up was conducted.
Brief report: family-cased group intervention for young siblings of children with chronic illness and developmental disability	Lobato & Kao, 2005 (USA)	43 well siblings (17 boys, 26 girls aged 4 to 7) of 40 children (25 boys, 15 girls aged 1 to 19) with chronic illness and developmental disability (ASDs 35%, ID 25%, physical disability 23%, medical disorders 14%, dual psychiatric disorder and learning disorder 3%) and 40 parents. Mean age of mothers 34, fathers 32. 100% Caucasian.	Families whose children were in the end stage of illness or who had been diagnosed less than 6 months before the study began were excluded	Uncontrolled case series, pre-post evaluation study. Study conducted in outpatient setting	Manualised collateral and integrated sibling-parent group intervention consisting of 6 90-minute sessions. Study duration not explicitly reported but assumed to be over 6 to 8 weeks as described elsewhere (Lobato & Kao, 2002). Skills taught: sibling knowledge, education about condition, identifying and managing emotion, problem solving, identifying strengths and needs, communication enhancement.	Sibling Knowledge of CI/DD via structured interview (Lobato & Kao, 2002); Sibling connectedness, Global functioning (via Pictorial Scale of Perceived Competence and Social Acceptance for Young Children; Harter & Pike, 1983); Child Behaviour Checklist (CBCL; Achenbach, 1991); parent satisfaction questionnaire	Sibling knowledge improved; cognitive and physical competence improved; satisfaction, completion and attendance was high; no other significant results. 3 month follow-up available for a subset of 17 families

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Brief report: family-cased group intervention for young siblings of children with chronic illness and developmental disability	Lobato & Kao, 2005 (USA)	43 well siblings (17 boys, 26 girls aged 4 to 7) of 40 children (25 boys, 15 girls aged 1 to 19) with chronic illness and developmental disability (ASDs 35%, ID 25%, physical disability 23%; medical disorders 14%, dual psychiatric disorder and learning disorder 3%) and 40 parents. Mean age of mothers 34, fathers 32. 100% Caucasian.	Families whose children were in the end stage of illness or who had beer diagnosed less than 6 months before the study began were excluded	Uncontrolled case series, pre-post evaluation study. Study conducted in outpatient setting	Manualised collateral and integrated sibling-parent group intervention consisting of 6 90-minute sessions. Study duration not explicitly reported but assumed to be over 6 to 8 weeks as described elsewhere (Lobato & Kao, 2002). Skills taught: sibling knowledge, education about condition, identifying and managing emotion, problem solving, identifying strengths and needs, communication enhancement.	Sibling Knowledge of CI/DD via structured interview (Lobato & Kao, 2002); Sibling connectedness, Global functioning (via Pictorial Scale of Perceived Competence and Social Acceptance for Young Children; Harter & Pike, 1983); Child Behaviour Checklist (CBCL; Achenbach, 1991); parent satisfaction questionnaire	Sibling knowledge improved; cognitive and physical competence improved; satisfaction, completion and attendance was high; no other significant results. 3 month follow-up available for a subset of 17 families
Modifying the 'Positive Parenting Program' for parent with intellectual disabilities	Glazemakers & Deboutte, 2013 (Belgium)	30 parents with ID (15 mothers, 15 fathers). Mean age 37.7. Mean no. of children = 3.48. Mean age of youngest child 5.73. Family demographics but are not reported on.	Parents with IDs as confirmed by Weschler Adult Intelligence Scale scores (WAIS-III, 3rd edition; Uterwijk 2000) with at least one child younger than 12 years of age	Uncontrolled case series, pre-post evaluation study. Intervention delivered in facility for assisted living	Modification of manualised group Triple P training for use with parents with IDs consisting of four group sessions lasting over 2 hours, three follow-up home visits, and between-session phone calls or home visits. Skills taught: developing positive relationships, managing and encouraging behaviour, teaching new skills, developing routine, problem solving.	Strengths and Difficulties Questionnaire (SDQ, Goodman 1997, 1999); Parenting Scale (PS; Arnold et al. 1993); Depression Anxiety Stress Scales (DASS; Lovibond & Lovibond, 1995); Client Satisfaction Questionnaire (CSQ; Sanders et al, 2000)	Significant reduction in parental report of child conduct problems, significant reduction in verbosity of parenting and maladaptive parenting overall, and a reduction in anxiety as well as overall depression stress and anxiety scores. No follow-up was reported.

Title	Study	Demographics	Inclusion/Exclusion	Study Design and setting	Intervention	Measures	Results
Family systems therapy with families with learning-disabled children: An intensive study of outcome	Eisenhauer, 1991 (USA)	3 families; 1: both parents aged 38, 11 year old ID child, 6 year-old sister. 2: mother 42, father 45, 2 twin boys aged 10, 1 with ID. Three older children no longer living in the family home. 3: mother 37, father 40, son aged 11 with ID, son aged 14.	Family systems therapy	Controlled trial. Non-treatment control group consisted of children in the same class as those in treatment group to reduce homogeneity. Intervention delivered in treatment centre room.	10 weekly 90 minute Family Systems Therapy sessions focused on problem-solving, and the here and now. Family characteristics and styles of relating were defined on a second by second basis according to a structured therapist manual. Individual counselling sessions followed completion of weekly structured tasks.	Revised Behaviour Problem Checklist; Devereux Elementary School Behaviour Rating Scale II; Weekly report cards; The Ericson-Rogers Relational Coding System	Improvement on academic performance in the treatment group relative to the comparison group, but most comparisons do not reach statistical significance levels. Slight decline in teacher's perceived achievement-compatible behaviours of the child. Improvements were found in parents perceptions of their child's behaviour improved throughout. Slight treatment effect was noted in parent relational dynamics, but not significant. Significant change in family relational dynamics are reported. Follow-up over 3 weeks, 60 days after final session.
Utilising BFT to help support the system around a person with ID and complex mental health needs: a case	Marshall & Ferris, 2012 (UK)	45 year old man, mild intellectual disability and schizophrenia and autism	None reported	Case Study. No control group present. Setting details requested from author.	Manualised Behavioural Family Therapy consisting of 11 hourly sessions delivered weekly. Typical BFT procedures were followed but not explicitly reported. Skills taught: problem solving, communication, expressing pleasant feelings, making positive requests, expressing unpleasant feelings.	Caregiver Strain Questionnaire, Family Functioning Questionnaire	Decreased parental strain; improved functioning. No official follow-up but some, community placement has continued until 30 months, where it would usually breakdown within 3.
An attachment-focused parent-child intervention for biting behaviours in a child with intellectual disability: A clinical case study	Mohamed & Mkabile, 2015 (South Africa)	13 year old with moderate ID, hemiparesis, and severe biting behaviour and her mother	None reported	Case Study. No control group present. Study settings are unclear	Non manualised, structured attachment-focused parent-child intervention and 'Theraplay' (Jernberg, 1984) consisting of three individual sessions with Mum, Eight 60 minute sessions of intervention following by a termination phase of two 60 mins sessions. Skills taught: awareness of others, acceptance and expression, reflective functioning, mentalisation	Systematic Interactions analysis using the Marschak interaction method (MIM) compiling observation and reflection	Mum more engaged in play, more confident in responses, play was more spontaneous. Mum less absent, lethargic and withdrawn, more energised and diverse. Joint tasks increased in duration and less mechanical. Physical proximity, direct contact, gentleness and sensitivity improved. Increase in warmth, affection and nurturance. Increased smiles from Mum and daughter. Improved mentalisation in mum. Observations supported by Mum's self-reflections and nursing staff. Decreased biting behaviour and improved relationship. Follow-up at 3 and 6 months.

Given the variety of participant difficulties, it is logical that corresponding interventions are also heterogeneous. Six of the studies involved variations on traditional systemic therapy models (i.e. behavioural family therapy, adapted family therapy, group family therapy, and family systems therapy). Three involved parent-child dyads exploring systemic dynamics and processes through play, one involved sibling-parent dyads. One study employed a parent training approach based around the well-established 'Triple P' programme, which was modified for intellectual disabilities and involved additional systemic elements. Lastly, one study explored a relatively new systemic intervention that involves the individual with an intellectual disability as well as the wider network of support resources, including other health professionals. Although several relationships were considered throughout the discussed research, most of the interventions were designed for use with parents and their young child with intellectual disability. This is problematic given the widespread and enduring impact that these difficulties can have on the system across the lifespan. Health care workers were only considered in one study, though no outcomes were collected to determine intervention effect with this group. Participants in the studies ranged from under 12 months up to 45 years of age, although parent ages were not specified in several studies. Participants across studies came from a diverse range of backgrounds from five different

countries around the world. By design, nine of the twelve included studies did not involve control group comparators, therefore the cause and effect of interventions is unclear.

As Graph 1 indicates, the overall quality of the articles included is poor. It is however important to note that the scoring tool used was tailored for the purposes of this review, therefore low scores may reflect strict criteria. The highest scores across the studies relate to the research question and participant characteristics. The research question was well defined, with specific outcomes explicitly explored in eleven of the twelve articles. A detailed description of participant characteristics was present in nine of the articles. None of the studies involved blind treatment assessment. This downfall likely relates to the type of research being conducted. Similarly, just three of the studies involved a control group. Without this, it is difficult to determine whether the observed outcomes are a result of a given intervention or simply an inevitable effect of change over time. Only five report on the management of missing data and few of the studies collected data from outcome measures that were specifically validated for use with people with intellectual disabilities. The results therefore must be interpreted with caution. The paucity of research involving intellectual disabilities makes it difficult to use measures sufficiently validated for this population. These factors contribute to the limited value of research overall.

Discussion

Systemic approaches are commonly employed for use with people with intellectual disabilities and their families. Research supports the theory behind their utility for this population (e.g. Daley et al, 2015; Fidell, 2000; Hashim, 2016), but this review highlights the distinct lack of empirical evidence-base to support its use. Though some researchers postulate the applicability of systemic principles for families including people with intellectual disabilities (e.g. Goepfert et al, 2015), these interventions are frequently adapted from their traditional format with no evidence base or clear rationale.

It is clear from the literature, and from this review alone, that the additional difficulties faced by people with intellectual disabilities and their families are varied, differing hugely from person to person. Certainly, intellectual disability is not a single disease entity, but a large group of heterogenous disorders with impaired intellectual abilities as their common denominator. Intervention adaptations cannot be standardised for use with all people with intellectual disability due to this diversity. It is more appropriate to tailor treatment to the needs of each individual system or family, which makes it very difficult to manualise and in turn, renders it difficult to evaluate. Indeed, families themselves are not a homogenous group,

therefore again it may be naïve to expect a one-size-fits all model to benefit this mixed group. It is possible that systemic therapies are beneficial for a proportion of this group with certain characteristics and not others, or perhaps just specific elements of certain interventions are beneficial for certain people. It is out with the scope of this review to examine each element of systemic work to determine which may be most helpful and for whom. Certainly, one of the main difficulties in completing this review was ascertaining whether the interventions used could be defined as systemic or if they were simply an amalgamation of several treatment approaches with some systemic elements. Relational shifts may also be reported even when no direct work has been conducted to target this. This could help to explain why mixed results are yielded when evaluating such interventions.

One of the other issues for research in this field, is that it doesn't always lend itself well to randomised control trials. This kind of research often requires large quantities of data, collected via objective outcome measures. The conspicuous scarcity of outcome measures, validated for use with people with intellectual disability often results in researchers using measures designed for neurotypical populations, which is likely to reduce their validity. Several of the present articles used unvalidated measures to base their conclusions on, which poses a concern around the value of that research.

Furthermore, the subtle and complex relational changes often sought in systemic work are not always captured by objective outcome measures. These changes are perhaps best explored through more qualitative methods such as observations and interviews that allow for more in-depth exploration into treatment effects, although predicted consequential changes in things like stress levels may be objectively measured to support a given hypothesis. Difficulties in implementing RCT based research with this population to evaluate intervention is also complicated by issues in blinding participants to their treatment group as people will be aware of beginning intervention or not. Although some efforts can be made to mask a control intervention by omitting therapeutic involvement, some would argue that it is immoral to have waitlist controls when families need intervention, and that engagement in any service, whether it involves psychological intervention techniques or not, may be considered therapeutic and helpful.

This review considers several different interventions used with people with intellectual disability, therefore it is not possible to draw conclusions about 'systemic therapy' in general. It is however interesting to synthesise some of the shared elements of these interventions. The most common elements were parent training components, skills development to support behavioural modification, and elements of work on family functioning such as identifying

unhelpful transactional behaviours, identifying the strengths and needs of the system, and improving relationships within it. Other commonalities include modules or sessions relating to problem solving, enhancing communication, and developing coping skills. It is perhaps not surprising that the most frequently reported significant changes related to improvement in the behaviours of the individual with the intellectual disability (5), parents or carers (3), and sibling behaviour (1). Five articles report on improved relationships, interactions or communication between family members, and two note improvements in the parental expectations or perception of problematic behaviour. Improvements in caregiver strain, depression and anxiety were reported in three studies, though one study found no change in parental levels of stress following intervention. Similarly, no changes were observed in relation to relationships (2), parental stress (1), parental expectations or perceptions of their child's behaviour (2), or the identified individual's behaviour (1) in some articles. In fact, two studies report on a decline in expectation and in behaviours post intervention. Because of the nature of design, it is not possible to extrapolate which elements are of most benefit to whom, although some studies did explore differences in outcomes according to mode of delivery, i.e. group, self-taught, and facilitated. The mode of delivery also differed in relation to participant involvement. Although the literature search criteria specifically

looked for systemic interventions, it is interesting that two of the presented studies involved no in-session work with the person with intellectual disability themselves. One study involved only siblings and parents of children with intellectual disability. The implication may be that it is of more benefit to work with the systems around these individuals, nodding to Freud's theory that people with intellectual disability cannot benefit from individual-focused, introspective therapy. However, the growing shift towards holistic, person-centred approaches to individual intervention may well necessitate systemic considerations given the inextricable, yet integral relational and potentially reinforcing dynamics of a system. Indeed, the whole is greater than the sum of its parts (Aristotle).

Limitations

Articles included in this review were selected from a pool available online, though attempts were made to source those that may not have presented through manual reference searches and through interlibrary loan requests. Included studies were also restricted to published and unpublished journal articles, no books or other literature were included. Unpublished articles were included following review, because their content was deemed relevant, however their lack of peer review is an acknowledged limitation.

Included articles were also restricted to those available in English, which may present publication bias.

Given the diversity of intervention approaches, determining whether the intervention was 'systemic' as previously defined posed a difficulty. Many included interventions involved 'the system', yet little focus was placed on the intricate dynamics or relationships between its members. Emphasis in a number of these interventions was on skill teaching and development or psychoeducation, rendering it less systemic, and more behavioural or informative. The relatively recent evolution in the development of traditional behaviour parent training first introduced by Griest & Forehand (1982) has led to increased focus on family systems, which makes the intervention model or approach more ambiguous. The diverse understanding of what systemic therapies must involve, makes the selection criteria of this review somewhat subjective. Similarly, few authors reported on the detail of the approaches involved in their studies. This too made it difficult to ascertain whether a true systemic approach was being investigated. Although interrater agreement account for this ambiguity to some degree, it remains a noted limitation. A lack of detail provided related to specific intervention procedures also complicates this discernment. The inclusion of other psychological intervention strategies alongside

systemic treatment contaminates the evaluation of systemic intervention in its pure form.

The review search is only as good as its terms. Although not specifically searched for, a proportion of relevant articles related to parenting interventions for those caring for people with intellectual disabilities. Had the search criteria included parents, or training, the results would have been vastly different. Similarly, parental involvement largely pertained to mothers. Although fathers were invited and participated in all but two of the studies, far fewer paternal figures engaged in interventions compared to maternal figures. Although this review involved participants across ecological levels, i.e. from academic and familial standpoints, none of the included studies collected outcomes from healthcare workers, or hospital staff. In order to serve justice to the model, assessment must be completed from multiple vantage points, which would allow exploration into widespread potential benefits.

Conclusions

This review identified mixed outcomes for the implementation of systemic therapeutic approaches for people with intellectual disabilities and their families or carers where there are additional difficulties with behaviour and/or mental health. Although the

rationale behind employing this approach appears sound, the evidence-base to support its utility lacks specificity, quality and power. It is possible that the paucity of quality research highlights the complexity in evaluating approaches of this ilk. However, more research is required to fine tune this approach to better understand the elements of intervention that effect change, and to better support systems.

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Behavioural Family Therapy to Support Adults with Intellectual Disabilities: Primary Caregivers' Perspectives

Running Title: BFT for Intellectual Disability

Key words: BFT; intellectual disability; family therapy, IPA

Word Count: 17,853 (excluding graphs and tables)

Abstract

Background: People with intellectual disabilities are at greater risk of developing mental health problems, often leaving carers at greater risk of stress and mental health deterioration. Behavioural Family Therapy (BFT) is a form of psychological treatment that has been developed to reduce stress within families.

Research Question: BFT is delivered widely across UK Intellectual Disability Services, yet little is known about its utility, or how it is received. This study aims to explore primary caregivers' experiences of being a BFT participant.

Method: Eight primary caregivers participated in an interview about their experiences. Interpretive Phenomenological Analysis was used to analyse interview data.

Results: When asked about their experience of BFT, caregivers reported their difficulties with openness, the challenges they faced related to engagement, and the factors that assuaged these issues. Caregivers also reported on the changes and adjustments that were experienced as a result of BFT. Improvements in communication and problem-solving skills, as well as increased insight and compassion between members, was a common feature of caregiver narratives. Interestingly, caregivers also unanimously discussed components of stress related to their caregiving role, and the impact that this stress

has on their lives. Three super-ordinate themes emerged; journey to acceptance and engagement in BFT, sense of progression and change, and caregiving in a challenging system. Psychological, analytical abstractions and extrapolations are presented in relation to overall intervention process and reception.

Conclusions: Results indicate that the well-evidenced benefits of BFT, identified in the psychosis and schizophrenia literature, are also perceived by caregivers of individuals with intellectual disabilities. Clinical implications and recommendations are described, which include promoting trust within the therapeutic relationship, emphasising the benefits of whole system engagement, and considering how current stress may impact on caregivers' ability to engage and support BFT intervention.

Introduction

The Scottish Government's 'The same as you?' strategy (2000), proposed to improve the community-based services available to people with intellectual disabilities, led to the closure of over 1000 long-stay hospital beds (Scottish Executive, 2000). Many people were discharged to their family home to be supported and cared for by relatives or paid carers. In 2011, 44% of adults known to local authority services with an intellectual disability, were living with a

family member (The Scottish Consortium for Learning Disability eSAY statistics report, 2012).

Although caring for someone with an intellectual disability can be a positive experience (Blacher & Baker, 2007; Chadwick et al, 2012), the bulk of research on caregivers' experiences focuses on the burdens and challenges associated with this role (Maes et al, 2003; Ben-Zur, Duvdevany & Lury, 2005; Chou et al, 2009; Hill & Rose, 2009). Evidence suggests that caregivers are at a greater risk of developing physical health problems such as headaches, gastrointestinal problems, sleep disturbances, and respiratory infections (Gallagher & Whiteley, 2012; Bédard, Koivuranta & Stuckey, 2004). Research also indicates that people caring for someone with an intellectual disability experience greater levels of stress (Emerson, 2003) than people in the general population. Due to the lifelong nature of intellectual disability, the stresses experienced by carers are correspondingly pervasive, in comparison to those caring for family members with more transient conditions (Glidden & Schoolcraft, 2003). Other factors implicated in increased stress levels include the acceptance of the intellectual disability diagnosis, loss related to the person with the intellectual disability not achieving certain milestones and aspirations (Hastings & Beck, 2004), the perceived vulnerability of the person with the intellectual disability, worries about future support provisions, and the transition from

child to adult services (Fidell, 2000). Furthermore, carers of someone with an intellectual disability are also at greater risk of psychological comorbidity, particularly depression and anxiety (Emerson, Robertson & Wood, 2004; Gallagher, Phillips, Oliver & Carroll, 2008). Caregiver burden is implicated as the link between caregiving and psychological distress (Bhatia et al, 2015), though the precise route to this elevated risk is unclear. What is known, is that the duration of caring responsibilities positively correlates with caregiver's experience of strain and psychological difficulty (Tsai et al, 2009; Seltzer et al, 2011).

Improvements in health and social care have led to increased life expectancy in the general population, including in people with intellectual disabilities (Emerson et al, 2012). Therefore, just as clients are living longer, so too are their caregivers. Consequently, the duration of caregiver and care-receiver roles are becoming increasingly elongated. Many individuals with intellectual disabilities continue to reside with their family at home well into adulthood (Braddock et al, 2013). It is estimated that 29,000 people in the UK with an intellectual disability are living with a family member over the age of 70 (Foundation for People with Learning Disabilities, 2010). Given that family caregivers form an integral part of service provision, it is essential that their needs and perspectives are

understood and addressed, to appropriately sustain this valuable resource.

A previous study exploring caregivers experiences of caring for a child with a range of disabilities reported on 5 emergent themes; 'stress of caregiving', 'negative impact on caregiver health', 'sharing the burden', 'worry about the future', and 'caregiver coping strategies' (Murphy et al, 2007). The authors concluded that interventions that address these issues will positively impact on caregiver health, wellbeing, and capacity to maintain their caregiving role. Research posits that families who can manage their stress proactively, are better equipped to assist other relatives to stay well (Kuipers, Leff & Lam, 2002). Research also suggests that families with good mental health are more resilient (Choi & Yoo, 2015), and therefore it is reasonable to assume that promoting mental health and reducing stress levels in caregivers as well as clients is important (Rapanaro, Bartu & Lee, 2008; Willigham-Storr, 2014) and likely to foster better outcomes.

Stress or illness affecting one member of a system is likely to have repercussions for all other members (Waters, West & Mendes, 2014; Oberle & Schonert-Reichl, 2016). People with intellectual disabilities, perhaps more so than those without, live within complex social systems, with dependence on an intricate and complex network of

carers, family members, friends and various services and agencies. Hence, caregiver distress is likely to increase distress in the individual with intellectual disability and vice versa. Poor caregiver health is also associated with recurrent hospitalisation for the client (Kelly & Hewson, 2000), and unwanted decisions to remove the client from their home (Llewellyn et al, 1999). The rationale for the utility of family interventions for people with intellectual disabilities is therefore clear, however very little research on the use of family interventions for people with intellectual disability exists to date.

Approximately 1.4 million people in the UK have a learning disability (Office for National Statistics, 2017). Though prevalence estimates vary from 3.9 to 97% (Buckles et al, 2013; Cooper et al, 2007), evidence indicates a greater incidence of mental health problems in people with intellectual disability compared to the general population (e.g. Emerson & Hatton, 2007; Cooper et al, 2015). One might assume that this would result in increased service provision for this population. However, research indicates that people with intellectual disabilities have been historically disadvantaged by a lack of access to psychotherapeutic approaches (Kroese, 1998; Bender, 1993). This is likely a consequence of the historical, yet unevidenced assumption that people with intellectual disabilities are more immune to emotional difficulties or impervious to the benefits of psychological intervention (Freud, 1904). Although many have since

disputed the presumed limited value of psychological intervention (see Waitman & Conboy-Hill, 1992 for a preliminary overview), this assumption has also contributed to increased interest into family interventions for people with intellectual disabilities.

Family interventions are a set of approaches that utilise a variety of strategies to reduce stress and burden; to improve the family's emotional atmosphere; improve problem-solving abilities; reduce expressions of anger and guilt; align patient and family expectations; and promote helpful change in family belief systems. Early forms of family intervention focussed on the modification of problematic behaviour in children, such as anxiety, nocturnal enuresis, and aggression (e.g. Wolpe, 1958; Williams, 1959; Risley & Wolf, 1967). Family-based interventions have since been adapted to inform parent training programmes and couples therapy (Baucom & Lester, 1986). Success in these areas triggered further exploration of its' application to mental health problems such as Dementia (Benbow, Marriott, Morley & Walsh, 1993), Bipolar Disorder (Miklowitz et al, 2007), severe mental illness (Campbell, 2004) and Eating Disorders (Eisler et al, 2007; LeGrange & Robin, 2017), all of which have shown promising results by improving things like communication and problem-solving skills.

BFT is a manualised, yet flexible form of family-based therapy, developed by Falloon (1982). It is used to support families that

include a member with enduring mental health problems. It is a skills-based intervention that promotes positive communication, stress management, and problem-solving skills (Campbell, 2001; Fadden, 2009). It typically takes 10 to 14 sessions to deliver (see Table 1), and involves information sharing with the client and their families related to the client's mental health issues, treatment, and experience. Families will work on recognising the clients' early signs of difficulty and will develop a clear plan to help the client maintain optimal wellbeing. Like other family interventions, the focus of BFT does not lie solely on the person with mental health difficulties, but instead addresses the needs and strengths of all family members as a group. BFT therapists require specific training in order to facilitate BFT intervention, which is provided by The Meriden Programme. This NHS Programme is the largest training and development programme of its kind in the world and is dedicated to improving access to family-based interventions.

Table 1: Generic BFT session outline (The Meriden Family Programme)

Meeting with the family to discuss the benefits of the approach
Agreement from the family to engage
Assessment of individual family members
Assessment of family communication and problem-solving skills

Review of the assessment on family resources, problems and goals
Meeting with the family to discuss how to proceed and how to establish family meetings
Information sharing regarding relevant mental health issues to reach a shared understanding
Work on early warning signs and relapse prevention. Development of 'staying well' plan
Helping the family to enhance and develop effective communication skills
Supporting the development of the family's problem-solving skills
Sessions to recap and booster previous work
Review and on-going support or discharge

The bulk of evidence for BFT in a mental health context relates to schizophrenia and psychosis (Berglund, Vahlne & Edman, 2003; Li & Arthur, 2005; Fadden, 2009). Pharoah et al (2006) found that BFT helped to reduce the stress and burden experienced by carers of individuals with schizophrenia. Reduced expressions of anger and guilt, resulting from engaging in this intervention, also helped to improve the emotional climate within the family, which in turn prevented relapse. A systematic review of early interventions for psychosis highlights that family intervention such as BFT, that focused on disorder education, enhancing problem solving skills, and

crisis management, helped to increase medication adherence, and reduce rates of relapse and hospital admissions (Bird et al, 2010). Recommendations now promote utilisation of BFT for psychosis and schizophrenia (NICE Guidelines for Schizophrenia and Psychosis, 2014; SIGN).

Although a large body of evidence supports the use of family interventions for helping families to cope with the experience of a relative with psychosis (e.g. Alpi et al, 2008; Addington, McCleery & Addington, 2005; Onwumere, Bebbington & Kuipers, 2011; Jones, 2009), one review of the literature indicated that only moderate benefits were achieved (Pharoah et al, 1999). Indeed, little is known about the specific ingredients that effect positive change (Pfammater et al., 2006). The reviewers note that better insight into the potential benefits of this intervention could be obtained via subjective reports of families who had received the intervention. Subsequent research exploring caregiver's experiences of family interventions for schizophrenia and psychosis indicates that they felt that it was "too little, too late" (Campbell, 2004). However, they also reported feeling empowered by an increased understanding of their relative's disorder, they felt benefit from reattributing unhelpful behaviours to the illness and not the individual, and that their communication skills improved as a result of the intervention (Budd & Hughes, 1997).

There are many similarities between the difficulties reported by

these families supporting individuals with schizophrenia and those supporting individuals with an intellectual disability, i.e. carer stress and burden, and feelings of frustration. It is therefore reasonable to consider whether families of individuals with intellectual disabilities may also benefit from these family interventions.

Implementation of BFT in intellectual disability services is a relatively new initiative, though preliminary research provides promising insight into its potential usefulness. Marshall and Ferris (2012) report on outcomes regarding the utilisation of BFT with the support system around a man with a mild intellectual disability, autism and schizophrenia. Sessions involved establishing relationships and boundary setting, information sharing about the client's history and diagnoses, enhancing problem solving skills, developing communication skills (expressing pleasant feelings, making positive requests, and expressing unpleasant feelings), and crisis prevention and maintenance plan development. There was no change in other treatments during the intervention period. Authors reported a reduction in carer strain, and improvement in functioning following intervention, observed in the client's mother. The functioning of the external support team also improved, and the client managed to maintain his community placement in Scotland for 30 months following the intervention, where they had previously broken down within 3 months. Hutchison et al, (2017) report on therapist

experiences of BFT for people with intellectual disabilities, which highlighted the value and benefit perceived as a result of the intervention. They also noted the difficulties experienced by therapists when implementing BFT, such as challenges engaging the family, inappropriate referrals, the duration and timing of the intervention, logistical obstacles, organisational barriers, and limited understanding of the intervention in clients and families. This resonates with previous literature on the implementation barriers (Lee et al, 2012; Smith & Vellman, 2002; Absalom-Hornby et al, 2011; Powell et al, 2013). A feasibility study (Anderson et al, not published) conducted throughout five Scottish NHS Health Boards, showed that of 76 families including an individual with intellectual disability referred to BFT, 45 did not complete treatment, with the highest attrition occurring during the engagement phase. This instigated interest into the perceived benefit and relevance of BFT for these families, which could be explored through investigating their experiences.

This study aims to explore the experiences of caregivers (of individuals with an intellectual disability) of being a BFT participant. As caregivers play an integral role in both caring for their relative, and in facilitating family intervention, it is important that their opinions on these experiences are sought. Caregivers are a powerful

source of information and often provide the majority of service provision for people with intellectual disabilities. It is therefore essential that their perspectives are considered when designing and implementing interventions. There have been no studies to date that have sought to explore the experiences of these caregivers in relation to their participation in BFT. Yet, as has been outlined above, their experience underpins this treatment's effectiveness, and therefore it is essential that they are explored. Exploration of experiences will not only help to ensure caregiver satisfaction, but research also indicates that participant attitude towards treatment can influence its outcome (Moradveisi et al, 2013) and that the invitation to offer their opinion alone, can make caregivers feel valued and appreciated (MacDonald, Sinason & Hollins, 2003). Furthermore, caregivers who perceive support to be helpful, report lower levels of stress, depression, and anxiety (White & Hasting, 2004). It is therefore important to explore what their experience of receiving support might tell us about their conclusions of its benefit. The link between carer and client well-being further substantiates this need. It is hoped that a greater insight into caregiver perspectives of being a BFT participant, may enhance our understanding of the issues they face, and how they relate to their experience of therapy. A better understanding of their experiences may also provide insight into issues related to engagement, attrition, service delivery, treatment

suitability and adaptation, as well as the specific considerations that therapists should hold in mind when delivering BFT.

Method

Design

Qualitative methods were employed to explore the experiences of eight primary caregivers of an adult with an Intellectual Disability who had received BFT as part of their routine Intellectual Disability service. Qualitative research focuses on a comprehensive inquiry of a small target audience, in this instance, caregivers, to explore how participants make sense of their experience (Patton, 2015). Qualitative research also allows the researcher to gain insight into the context and systems that influence and contribute to their interpretation of their experience (Crowe, Inder, & Porter, 2015). Semi-structured interviews were conducted, which gathered detailed interpretations of participants' experiences. Deductive approaches were used to build upon existing literature and to explore preconceived objectives in light of gaps in the evidence base. Inductive approaches helped to explore innovative information in this evolving field of research, based on the unique data provided by this group of caregivers.

Recruitment

A convenience sampling approach was employed, as participants were recruited based on the availability of participant data from a centralised database. Ethical approval was granted from the West of Scotland Research Ethics Service and from the research and development department in each of the three Community Intellectual Disability Teams involved across Scotland. The Team Leads of each service assisted with recruitment for their site by consulting the database of clients who had received BFT and then identifying those who had completed at least 5 sessions and had progressed past the engagement phase. Primary caregivers, as identified by relevant clinicians, were then highlighted. A recruitment email was also sent to all relevant clinicians (Appendix 6), who could also identify eligible primary caregivers who met the inclusion criteria. Eligible caregivers were then contacted by their local Intellectual Disability Team to ask whether they would be willing to take part in an interview about their experiences of being a participant in BFT. Candidates were also provided with a Patient Information Sheet (Appendix 7), which described the elements of participation involvement. These sheets contained a note of interest form, which would be returned including contact details and an indication of willingness to participate. The first author then contacted interested candidates in the manner indicated by their

note of interest form, to answer any questions they may have, and to arrange the interview. Prior to interview, the first author went through the information sheet with participants, reiterating the purpose of the study, what it would involve, and their right to withdraw from the study at any time. If there were no concerns, written consent (Appendix 8) was obtained, and the interview commenced.

Participants

Eight participants who met the inclusion criteria (Table 3) were interviewed. Participants could be excluded from the study if it was deemed inappropriate or unsuitable by their

known clinician. All identifiable information had been altered to ensure anonymity. Six participants were interviewed individually, and two, Pete and Diane, were interviewed as a couple, as both were considered by themselves and their clinicians to be primary caregivers and indicated a preference to be interviewed together. Participant characteristics are detailed in Table 2. Participants included five mothers, two fathers, and one male foster carer, who had been caring for the client for approximately 3 years at the time of interview. Participants were from a range of socioeconomic backgrounds across three health boards, NHS Lanarkshire (3), NHS Lothian (2), and NHS Ayrshire and Arran (3). One further participant

noted interest, however it transpired during the interview that criteria were not met. Diane, Pete and Moira have cared for the client in the family home since their birth. Kerry's daughter (client) was moved to supported accommodation due to difficulties with caring for her at home. Mary and Andy's daughter (client) lives independently near their home and receives regular support. Christine's daughter (client) chose to move out of the family home into sheltered accommodation for the homeless approximately one year prior to the interview.

Table 2: Participant Characteristics

Caregiver (relation-ship to client)	Age Range	Employment	Relation-ship Status	Client Living Arrangements	Stage of BFT
Andy (father)	60-70	Retired	Married	Lives nearby and receives daily support from parents plus external support	Completed
Derek (foster parent)	40-50	Foster Carer	Single	Client supported in same residence	Ongoing
Mary (mother)	60-70	Employed	Married	Lives nearby and receives daily support from parents plus external support	Completed
Christine (mother)	60-70	Employed	Single	Resided in homeless shelter at time of interview	Ongoing
Kerry (mother)	40-50	Employed	Married	Supported in own accommodatio n nearby	Did not complete
Moira (mother)	40-50	Employed	Married	Client supported in same residence	Completed
Pete (father)	40-50	Employed	Married	Client supported in same residence	Completed
Diane (mother)	40-50	Employed	Married	Client supported in same residence	Completed

Table 3: Participant Inclusion Criteria

Primary caregivers for an adult with an intellectual disability and additional mental health problems and/or behaviour that challenges, who have accessed the Intellectual Disability Service in one of three health boards (NHS Lanarkshire, NHS Lothian, NHS Ayrshire & Arran).
Primary care givers who have attended a minimum of 5 BFT sessions and progressed beyond the engagement phase of intervention as part of the service user's routine clinical treatment. They may still be engaged in intervention at the point of interview.
Primary caregivers who have consented to take part in the study
Primary caregivers who speak fluent English
Primary caregivers aged 16 or over

The Interview

An interview guide (Appendix 9) was developed to guide the semi-structured interview. Questions were developed in light of those used in another study, published as part of the same, larger project (Hutchison et al, 2017). Open questions were used to allow caregivers to describe their experiences of being a BFT participant, with minimal restriction or guidance on their response. The initial question was intentionally vague; 'you and your family have done BFT, what was it like?' This allowed more freedom for any element of

their experience to be raised and explored. This was in keeping with the exploratory nature of this study's research aims and existing guidelines (Creswell, 2003). Additional prompts were used for caregivers who struggled with this lack of guidance and were chosen to provoke further consideration of their general experiences of BFT, as well as the elements perceived to be helpful and unhelpful. Questions were followed up with further enquiry, both general and specific, into their experiences to enhance the researcher's understanding and to encourage further reflection from the caregiver. Asking specific questions derived from the initial experience being described by the caregiver and from existing literature in the relevant area is a technique known as 'funnelling' (Smith & Osborn, 2008). This can help to elicit elaboration of interviewees description, reflection, and interpretation of their experience. The interview guide helped to ensure that the same lines of enquiry were investigated with all participants (Patton, 2015). However, each caregiver raised different issues in response to interview questions, therefore each interview uniquely deviated from this framework to encompass a variety of perspectives. It was important that rapport was established between the caregiver and the interviewer (Wagstaff & Williams, 2014), therefore interviews were preceded by more generic conversation. This helped to alleviate any worries or anxieties held by the caregiver, promote a trusting

alliance, and to enhance the quality and depth of information shared. Interviews were also conducted in caregiver's chosen location, which in all cases was their home. This also helped to ensure optimal comfort to promote ease of discussion. Six of the participants also received BFT in their home, therefore this setting helped the researcher to gain a better understanding of their experience (Creswell, 2012). Interviews were conducted by the first author throughout 2018 and 2019 and lasted between 43 and 99 minutes, with a total interview time of 7 hours and 46 minutes.

Procedure

The first author presented this study proposal to one NHS Community Intellectual Disabilities Team, to obtain feedback on methodological issues before initiation. Following feedback, participants were contacted by the Team Lead of their local service to determine their interest in study involvement. Interested participants consented to their information being shared with the first author who then contacted them to arrange the interview and clarify aspects of the procedure that remained unclear. Interviews were recorded using an encrypted recording device and transcribed verbatim, though all identifiable information was anonymised. The term client is used to refer to the individual with an intellectual disability. Alias names were used for all involved participants. Anonymous, descriptive language was used where other names were

mentioned, which detailed their relationship to the caregiver or the client. The following conventions were used:

… Words omitted

[ ] Additional explanatory information/translation

- Break in sentence

Full stops were used to denote short breaks and pauses. Longer pauses were described as so in brackets within the transcription. Emphasised words were written in bold, and additional descriptive information was documented in brackets within the transcription. Additionally, notes of interest were made following each interview to aid future recall of the interview and to enhance the documented detail of the interview environment and process. Notes included any thoughts and feelings experienced by the author during the interview, significant interruptions such as others entering the interview room, and any other relevant details such as tearfulness, laughter, or changes in body language.

Analysis

Interpretive Phenomenological Analysis (IPA) was utilised, following the flexible guidelines proposed by Smith & Osborn (2008). IPA was used to extract information about the meaning assigned to caregivers' experiences and to ascertain key themes across

participant accounts. IPA involves an elaborate interpretive process that explores the personal perspectives of each participant before generalising any elements of their lived experience (Thompson, 2018). IPA was chosen as its emphasis is on each individual experience and the meaning that they assign to it (Smith, Flowers & Larkin, 2009) from their unique perspective. It is specifically appropriate when exploring important or significant events in participants' lives, therefore it was useful to consider the significance that participants assigned to their experience of being a participant in BFT. The research field of intellectual disability is in its infancy compared to that of adult mental health, and much remains to be understood. Experiential exploration is therefore an appropriate means of gathering initial data to begin developing hypotheses. A growing evidence in other populations provides a top-down rationale for the use of BFT for families including someone with an intellectual disability, but less is known in relation to the bottom-up rationale, informed by recipients' experiences of it. The IPA approach is also idiographic in nature, which involves a detailed analysis of a case, therefore lends itself well to smaller sample sizes (Eatough & Smith, 2017).

Detailing the variety and intricacies of individual experiences whilst also exploring shared experiences amongst participants requires and promotes creative thinking to reserve meaningful insights from both

(Thackeray, 2015). The interviewer and interviewee were considered to influence the interview process, how it was experienced, and how it was interpreted (Gadamer, 1960/1990; Harrison, MacGibbon & Morton, 2001). Indeed, we do not enter social situations without reference to previous experience, knowledge, and understanding of similar situations. Participant language is understood as a significant means of recounting the meaning of their experience, and their experience is understood to be shaped and influenced by a number of factors such as age, gender, health, and various cultural norms and practices. Similarly, the researcher is subject to prevailing factors, and forms an integral part of the world they strive to recount. The author had no previous relationship with any of the caregivers involved in this study and had not been directly involved in the development or delivery of BFT for this group. However, all authors undoubtedly reserve a variable amount of bias when engaging in research, which influences the process (Hunt, 2010). IPA involves a dynamic, 'double hermeneutic' process, in which the researcher plays an active role in making sense of participants' interpretations of their experiences, while participants simultaneously attempt to make sense of their own experiences. This necessitates effort from the researcher to acknowledge, and where possible and appropriate, disengage with preconceptions and prior assumptions (Dahlberg, 2006; Smith, 2007). Some assumptions were however necessary,

such as the assumption that caregivers would provide an honest description and interpretation of their experiences of being a BFT participant. The first author kept a reflective diary to ensure that any values, assumptions and beliefs related to the study were acknowledged in terms of their potential impact on data analysis. Reflexivity can support an understanding of how the researcher's own experiences and perceptions of the world can impact the research process (Morrow, 2005). Awareness of one's own potential influences on the research process allows the researcher to 'bracket' these biases, which allows them to be more receptive to the data in the present moment (Moustakas, 1994).

Each interview was transcribed and listened to several times in turn before being used within comparative analyses of all participant data. Transcripts were annotated with the author's thoughts, interpretations, and any other significant notes. With each exposure, the author became more aware of theoretical and analytical connections between comments, as well as the meaning behind the presentation, language and tone of the interviewee during the interview. Emerging themes were subsequently identified, abstracted and categorised according to content, or other theoretical connections, which considered the meaning assigned to their comments. The original transcript was referred to throughout this process to avoid any inappropriate interpretations of responses from

being abstracted. Emerging themes were listed chronologically, to allow easier investigation into analytical and theoretical links. It is not unusual for participants to raise unanticipated issues in response to the interview questions, or indeed to the interviewer themselves. The process therefore involves careful conceptualisation of these issues, which holds psychological concepts and theory in mind, whilst maintaining fidelity to what was actually said and meant by the participant. The process involves intensive fluctuations between inductive and deductive stances. Clusters of similar themes were then categorised to form a list of super-ordinate themes. Each therefore has a super-ordinate theme and supporting extracts from the transcription attached to it.

Every effort was made to ensure that knowledge of themes from previous interviews did not taint interpretation of future data. Although some researchers welcome this contamination, the intention of this study was to explore the breadth of experiences of BFT as well as the commonalities between them. A second and third researcher were recruited to assist with a proportion of the analysis. This triangulation involved each researcher independently reading a portion of the transcriptions to corroborate and validate the author's coding of themes and interpretation, as recommended by Smith & Osborn (2003). This helped to avoid any bias and ensure a more

accurate interpretation of the original transcript. See Smith, Flowers and Larkin (2009) for further detail on the process of IPA.

Results

Three super-ordinate themes and nine sub-themes emerged from the analysis, as summarised in Table 3. Although these themes are presented as distinct, overlap between some themes was apparent, particularly in relation to more general or overarching themes such as blame, or methods of coping.

Table 4: Super-ordinate Themes and Sub-themes

Superordinate theme	Sub-theme
Journey to acceptance and engagement in BFT	1a) Deconstructing barriers
	1b) Paving the way: the foundations of engagement
	1c) Bumps in the road: challenges for BFT
Sense of change and progression	2a) Hope versus hopelessness
	2b) Therapeutic guidance and benefit
Too stressed to reflect: Caregiving in a challenging system	3a) The battle for support
	3b) Sense of duty to protect
	3c) The impact of the caregiving role
	3d) "Who's going to fight for him?": The future beyond my existence

Theme 1: Journey to acceptance and engagement in BFT

When asked what BFT was like, all caregivers identified important components regarding their ability to accept and engage in the intervention. Three relevant sub-themes emerged. Extracts that encapsulate the essence of each theme are presented.

Deconstructing barriers

Caregivers expressed that they were initially unsure of what therapy might entail, or what would be expected of them, which evoked anxiety related to engaging in BFT. Many conveyed a fear of openness due to worry about how BFT therapists may perceive them. To shield themselves from negative judgement, embarrassment or shame, caregivers described developing protective barriers to keep others at a distance. Therefore, discussing personal topics, usually concealed to those outside, and often to those within the family too, was an experience that some caregivers found unusual, uncomfortable and exposing. Christine captures this sentiment in the following excerpt.

Christine: it's [BFT] not for everybody, nut. Nut [no. No]. She [client's sister] tried it. She did try it, but (pause) it's not for her…because she's a private lassie…keeps everything to herself

Christine emphasised her other daughter's attempts to engage, revealing an understanding that BFT can be challenging, and perhaps intolerable for some. She justified and accepted her daughter's disengagement by conveying an assumption that BFT requires participants to sacrifice their privacy. Andy elaborated on this idea by suggesting that BFT may feel invasive. Yet, he disclosed a familiarity with the feeling of transparency due to continual input from services.

Andy: If invasiveness is what it is, then we've had people in our life since [client] was born

Andy's language indicates ambiguity about the appropriateness of the word 'invasiveness' to describe BFT but highlights an appreciation that this is a possible interpretation for others. His reflection on what he perceived as incessant input, was apathetic, indicative of someone who has resigned themselves to experienced transparency and who has become desensitised to the impact of potential judgement. However, fear of judgement permeated most caregiver narratives, which often derived from feelings of embarrassment and frustration with being misunderstood. This was apparent in relation to their caregiving competence and was often associated with their sense of helplessness in managing client behaviours. Caregivers had to embark on a continuous, challenging journey of adjustment to the

role of caregiver for individuals who are likely to possess different characteristics and needs from those in their existing network.

Diane: You could mind read them saying 'oh see if he [client] was mine he would get that' and as he gets older and his behaviour's the same, it's harder to have an excuse or a reason to explain to somebody…It can be embarrassing. It can be. Because of things that he says and he does

Interestingly, Diane's belief about the judgements of other parents in her description, did not develop from verbal interactions with them, but originated from her own fears of not being a good enough caregiver, or from 'getting it wrong'. These experiences shaped her conceptualisation of negative public perception and led to increased efforts by her to maintain privacy.

Diane: Mum [client's grandmother] would say 'how did things go?' and I wouldn't go into detail cos it would just…go straight over their head…I genuinely think they [grandparents] were quite embarrassed to start with…it took them a lot of years to accept him [client]

Here Diane demonstrates her need for additional adjustment to the evolving reactions of those around her, who are simultaneously navigating their own adjustment to the client. By withholding information from others and creating a barrier to protect her from criticism, she ameliorates the adjustment process and manages her

fear of judgement. It is understandable given these negative interpretations, that sharing thoughts and feelings with BFT therapists might be challenging for caregivers, not least as it requires them to let go of their coping mechanisms by dismantling established barriers. For some, this process proved extremely challenging as it involved exposing themselves and taking the risk of being negatively judged. Indeed, Kerry chose to preserve her defence by electing not to divulge some information with therapists.

Kerry: we didn't want…that [issue] dealt with...outsiders [BFT therapists] basically

Many caregivers reflected on the offer of psychological intervention. Often caregivers wanted support, but expressed reluctance to ask for it, accept it, or admit that they were struggling. Moira's quote illustrates how she perceived the offer of BFT as an indication of her parental ineptitude.

Moira: we would think 'are we failing here? Are we failed? Why are they coming in because we're actually good as a family?'… you feel vulnerable so we are looking for like 'please help us, but don't make us feel bad'

Like Diane, Moira's negative appraisals related to assumed criticism of her parenting. Her use of plural pronouns indicates that she believes these sentiments were shared by others in her family and

perhaps by others receiving BFT. Moira laughed slightly following this statement, indicating her own sense of irrationality for considering that therapists might seek to make her feel bad. Nonetheless, her quote exposes this concern. In order to let down her guard, Moira wanted reassurance that support was acceptable, positive and appropriate. In order to accept and believe these reassurances, she required empathy, understanding and validation of the challenges she faced. This was particularly important due to the often-invisible nature of intellectual disability, and the resulting effect that has on the caregiving pressures and expectations of others who may not be able to observe the challenges involved in the caregiving role. The expectations of others to manage as 'normal', can contaminate their own coping expectations and lead to increasing pressures to manage without support. The following quote illustrates how validation refuted some of the negative self-attributions that developed as a result of Moira's struggles.

Moira: I think it's an acknowledgement of … you're not bad parents, you are struggling with a situation that exists, it's not a figment of your imagination … it let us know that we weren't the only ones that were facing this problem … you expect that, you know, you will function (pause) and everybody else seems to do it so … we'll be the same

Underpinning the caregivers' capacity to let down their guard, was their ability to trust their therapist. Caregivers unanimously articulated a need for trust within the therapeutic relationship in promoting openness. Trust promoted acceptance and engagement in BFT and moderated worries about negative judgement. The quality of trust varied between caregivers depending on their unique needs and experiences.

Mary: if you can trust that person [BFT therapist] who's with your daughter then you can relax a wee bit

Like many, Mary also developed a vigilant and protective stance in response to her sense of the client's vulnerability. She assumed a protective role, remaining alert to possible threat at all times. Trust in the therapist relieved her of this duty by providing a safe, supportive environment, whereby the therapist and caregiver's goals and expectations were aligned. The journey from protectiveness to trust allowed Mary to invest more energy and attention on BFT sessions.

The significance of trust for Moira however, related to trusting that her investment in BFT was worth the time, effort, and emotional turmoil associated with accepting support. Her use of the word faith in the following extract has connotations of a religious ilk, as though for her, there were no tangible or objective reasons for her to expect that BFT would be helpful, but her desperation and need to believe

that it would be, superseded any doubt. This faith, or hope permitted Moira to fully engage with BFT, despite initial difficulties and discomfort.

Moira: just have faith that you know that they've got the skill and the knowledge to- to be appropriate for you

Other caregivers highlighted the benefits of knowing the therapist prior to intervention, which fostered trust within the relationship and alleviated fears of judgement.

Andy: we kinda trusted [therapist] and we knew her, so we were probably more comfortable

Andy's use of ambiguous language such as 'kinda' and 'probably' indicates that a level of discomfort and caution persisted despite acknowledging the benefits of the established therapeutic relationship prior to BFT initiation. This highlights the difficulties that caregivers have in deconstructing their protective defences, likely due to the evolved, habitual nature of these coping mechanisms.

Paving the way: the foundations of engagement

Caregivers unanimously referred to the necessary prerequisites for engagement in BFT. The importance of effort and commitment in order to reap the benefits of BFT was highlighted by most caregivers. Indeed, many indicated the necessary duration and level of

commitment as a possible obstacle to engagement. Timing and caregivers' sense of readiness to accept and to contribute were also highlighted as crucial elements. For Derek, this meant managing other risk factors before he fully engaged.

Derek: we started doing the BFT and then there would be an incident so they would change the BFT to accommodate what's been happening

Derek highlighted the continual stress and risk experienced in his caregiving role. His need to keep the client safe overruled the need for BFT, and although he appreciated the flexibility of the therapist, Derek implied that risk and BFT are mutually exclusive. In other words, Derek could not fully commit and engage in BFT whilst managing current risk. The recurrent nature of these incidents reduced the likelihood that prolonged equilibrium could precede BFT initiation, which disrupted his ability to engage. Conversely, other caregivers proposed that their sense of crisis enabled acceptance of support. Moira felt unable to request support before hitting 'rock bottom'. Pain and suffering therefore became her motivators to engage.

Moira: I think if they'd come in maybe before that it might have been more difficult. I think we were in a state of readiness to accept and

listen and- and act … we needed help, you know, it was like, I cannae do this, you know, it was too much

Moira's use of pronouns in this quote suggests a communal acceptance of, and need for, support that she felt resulted from her personal struggle to cope. A united sense of requirement facilitated commitment and engagement from all members of her family. Indeed, commitment from other family members was important to many caregivers. For some, it was deemed appropriate to exclude other members, often siblings, from participating. Kerry felt that input from the client's siblings was unnecessary, as difficulties were ascribed solely to the client. Unsurprisingly, blame focussed exclusively on the client, led to reduced involvement and commitment from other members.

Kerry: My son [client's brother] wouldn't have done it anyway … he wouldn't want to do it. He wouldn't do it because he didn't feel that there was anything wrong (pause) in his part

Others however, reported on the value of engagement from the entire family, based on an assumption that resolving family difficulties is everyone's responsibility. Andy's quote demonstrates how he felt that without the input from everyone, BFT would not work.

Andy: if there's a 'cannae be bothered' link anywhere in the chain then you're … you're busted.

This sense of shared responsibility was mirrored by other caregivers, who agreed that the purpose of BFT was not just to provoke change in the client's behaviour, but the behaviours of their surrounding system too. Caregivers like Moira who adopted this stance, were more motivated to engage, and reported a more holistic benefit from their engagement.

Moira: we had all been listened to and…all our behaviours were adapting

Bumps in the road: challenges for BFT

All caregivers implied that tailored therapy to their family's specific needs was essential to promote engagement and optimal benefit. The importance of adapting information and materials to the needs and abilities of the client specifically, was ubiquitous and perceived positively.

Mary: every week [therapist] would bring … illustrations that were very good at explaining, particularly for [client]

However, despite the recognised advantages of appropriate adaptation in terms of client engagement, many also conveyed a

sense that the client's intellectual disability either prevented or moderated potential engagement and benefit from BFT. The speed of information processing was highlighted by many caregivers who could become frustrated, particularly when encouraged changes were minimal or slow to materialise. Derek's frustrations were often directed at the client, thereby reducing his empathy and aggravating his exasperations towards him. Apportioning blame solely to the client in this way reduced his belief that engagement in BFT could lead to any beneficial change.

Derek: the problem- it's not the course, it's [client] … the problem is getting [client] to participate. Sometimes he canna be bothered

In the following extract, Pete demonstrates how other caregivers mentally partition their loved one's core personality from the difficulties they pose. By externalising these factors from the client and directing his frustrations at the difficulties themselves, Pete's capacity to tolerate and empathise with problematic behaviour increased. Although his method of coping did not necessarily lead to a reduction in problematic behaviours, it did lead to perseverance of BFT. Consequently, BFT gave Pete a more positive, proactive orientation towards the future, as opposed to a reactive and hopeless one.

Pete: [client] really didn't put much into it … it wasn't because of what they [therapists] were doing, that's just his autism…puberty and hormones on top of the autism and the learning disability…it's not helped…he can't control it…we were getting more out of it than he was

Interestingly, many caregivers implicated the clients' own self-concept as an obstacle to accepting and benefitting from BFT. Client's perceived to hold a negative view of their intellectual disability, perhaps due to experiences of stigma, bullying and denigration, often rejected their diagnoses and the subsequent support that was offered.

Mary's conceptualisation of the client's self-worth illustrates her difficulty in encouraging her daughter to accept and engage in BFT. It also demonstrates an understanding of the client's resistance to support, which represents a universal challenge for all paid staff supporting individuals with intellectual disabilities.

Mary: She [client] says, 'I feel like her in pretty woman. It's as if I'm a prostitute'. See that's her perception of it 'cos somebody has to get paid to…to spend time with her

Andy felt that the clients' sense of self led to resentment and resistance to BFT. He believed that the client's acceptance of her

diagnosis would lead to better outcomes and overall wellbeing for his whole family, and was therefore frustrated by the client's disavowal.

Andy: [client]'s not very accepting of her situation, which is what generates some of the difficulties we've had to face

Andy and Mary made frequent reference to the client's rejection of words related to her intellectual disability, that held negative connotations for her, e.g. carer and disability. With the support of BFT therapists, a shared language was developed, which could be accepted and adopted by everyone involved. Although this was a continuing process that did not aim to shift the client's attitudes or feelings towards these words, Andy and Mary both reflected on how that led to a reduction in emotional outbursts in the client.

Theme 2: Sense of Change and Progression

There was consensus amongst caregivers, that BFT was useful, and had improved their day-to-day lives. Many reflected on specific changes attributed to the intervention, whilst others reflected on overall benefit. Although a sense was conveyed that not everyone would benefit, many caregivers made inferences about the necessary conditions that could support progress and change.

Hope versus Hopelessness

The pervasiveness of intellectual disability was a common feature of caregivers' descriptions of their difficulties, which conveyed a flavour of hopelessness for some. Andy's reflection on the permanent nature of intellectual disability and the persistence of specific difficulties, despite efforts to improve them, elicited a sense of hopelessness as well as acceptance.

Andy: it's an ongoing process and it'll be a lifelong process…she's not got the flu and it's going to be better next week, you know. She's got a learning disability for the rest of her life…it is what it is and what this [BFT] has helped us to do is make the most of it. Make the best of it

Here Andy accepts the chronic nature of intellectual disability and the continual challenges that are likely to arise. His way of coping with this reality, is to positively reappraise his difficulties. His expectation of BFT is not to 'cure' the client of their disability, but to support effective adjustment to the situation as it is. In contrast, other caregivers described a palpable sense of feeling stuck or trapped in relation to the chronic nature of intellectual disability. They expressed a limited sense of agency or ability to improve things as a result. Some caregivers attributed this lack of change to their

own failures, which perpetuated their sense of hopelessness as well as helplessness.

Christine: we're in a situation that sometimes we cannae (pause) we cannae get out of it

Christine's appraisal of her circumstance illustrates her sense of hopelessness and helplessness. Christine was attending BFT sessions at the time of interview, yet she implied that her continual efforts of improvement felt fruitless, resulting in a sense of 'giving up'. Indeed, she retracted any element of hope encompassed by the word 'sometimes' and replaced it with a concrete, negative statement that embodies hopelessness.

For some caregivers, participation in BFT helped to foster a sense of proactive agency and direction that promoted hope. The collaborative and educational nature of sessions gave clients and caregivers the opportunity to learn and practice the necessary skills to manage problems and communicate more effectively. Some caregivers felt empowered, that with these skills, improvement could occur exponentially, and that client independence was a possibility. It is likely that positivity and optimism did not abolish their fears about the future, but having some hope enabled them to tolerate and adapt to them more effectively.

Certainly, many caregivers disclosed a shift in mindset following BFT, from a negative and frequently hopeless one, to a more positive and hopeful one. This transition allowed caregivers to redirect their energy and attention away from worries about what might happen in the future, towards what they do have influence over in the moment, including BFT engagement.

Andy: what I've learnt through this as well is (pause) there's no point in being morose … Take your positives from this process and work with them. Not all the negatives…we don't get all gloomy about it

Andy expressed an understanding, almost empathy with those who are morose about their circumstances, indicative of an individual with shared experience. Andy's quote portrays a journey of learning about his own maladaptive coping strategies, which led to a change of mindset towards a more positive perspective. Like other caregivers, Andy acknowledges the morbid and depressing reality of ongoing difficulty, but has learned with experience, and the support and guidance of BFT therapists, that persistent efforts to change the unchangeable, or maintaining a negative focus, only leads to frustration and more upset. There was a sense from some caregivers that a positive approach to their circumstance was a superficial way of masking or even undermining the severity of their reality, as acknowledged in the following extract.

Andy: but…don't get me wrong, sometimes…we bang our heads for twenty minutes and then we're fine…sometimes I think you need the wee (pause)…it's no a reality check, it's not a reality check it's (pause) it's just a…it's not a sadness check either but it's a (pause) frustrated for [client] check

Andy projected acceptance, yet not resignation, of the negative aspects of his caregiving role as part of their journey. He emitted a sense that persistent optimism and hopefulness may invalidate the severity of distress and difficulties faced by the client and those around them. Again, this highlights his consideration and understanding of their experiences. Those who acknowledged and accepted the negative, hopeless elements of their role, yet made a conscious effort to focus on the positive, hopeful aspects, appeared better equipped to engage in BFT and more aware of their progress.

Therapeutic Guidance and Benefit

Caregivers consistently described the positive effects they attributed to BFT. They described becoming more tolerant and understanding of each other through open discussions about their emotions, instigated by the intervention. They also reflected on how the client gained capacity to consider their actions as well as their consequences on the family, before acting or reacting. Referenced benefits most

commonly included improved communication, increased consideration of others' feelings, reduced anxiety, and a more positive outlook on life, which correspond with the overall aims of BFT. These reflections also indicate some of the issues causing difficulties prior to input, most notably difficulties with communication, problematic relationships, a lack of family unity, and feeling unable to cope with, or control, challenging situations.

Mary: I would say generally it did improve relationships. Not that we didn't have good relationships in the beginning, but it would make her [client] stop and think…and us as well, as parents, 'cos you do tend to forget that her perspective is totally different

In emphasising the strength of her bond with the client prior to BFT, Mary exposes her evolved defensiveness and negative assumptions about the perceptions of others. She also reveals a supposition that relationship difficulties were a consequence of the client's lack of consideration. Throughout Mary's interview she retracted or softened statements about the client, where she believed they could be perceived in a negative light by the interviewer. Certainly, in this quote she continued to say that adjustments were also being made by her and her husband, thereby diffusing any culpability to them too. This redistribution exemplifies how caregivers and clients

learned to be more considerate of other's feelings and more accepting of the need for all components of the system to engage.

Many caregivers reflected on the opportunity that BFT provided for the family to bond and develop together. This shared goal stimulated a sense of connectedness, thereby enhancing and strengthening relationships within the system.

Moira: I think [client] felt part of- we were all together in it, it wasn't just focusing on [client] … they want to help you all, do you know what I mean? So it was that kind of keeping us all as a wee unit

Mary's quote implies an assumption that BFT exists predominantly for the client. However, she repudiates this assumption by affirming her sense of togetherness with her family. Mary's narrative emits a fear of familial dispersion, perhaps due to her sense of strain on these relationships. For Mary, BFT help to solidify family bonds, thereby alleviating these worries.

Caregivers credited BFT for providing a platform whereby they and the client could express their feelings in a safe environment and in a helpful manner. Some suggested that BFT 'gave them a voice', which indicates a sense that they were previously silenced. Christine noted how her own style and the client's style of communication has changed since completing BFT.

Christine: we [Christine and client] used to shout at each other for the least wee thing. But now (pause) we just sit and talk – 'what is your problem?' we can deal with it if you tell me, if we talk about it…that's the kind of things we've been doing now

Christine's reflections on this change incorporated a shared responsibility for their previous maladaptive communication, as well as recognition that their reactions may not have been justified. She attributed difficulties in resolving issues to a lack of openness and discussion, and radiated hope that by challenging this tendency, solutions could be sought. Although an underlying assumption that problems ultimately belong to the client was presented, Christine demonstrated a willingness and acceptance to share and support the client with her struggles. For Christine and other caregivers, BFT gave them the tools for communication that helped to break down communication barriers and enable productive discussions and problem-solving opportunities. Similarly, Moira reflected on the benefit of breaking down barriers but the importance of guidance from the therapists to support this.

Moira: we were kind of weak at that point, we were vulnerable and we kind of opened up a bit…so we were kind of led through it…we had actually been led. I think that's- it's like the strength

Moira explicitly exposed her sense of vulnerability, but suggested that the strength and guidance of the therapists evoked strength and power in her and her family. As aforementioned, trust is an integral ingredient in promoting openness, yet Moira also demonstrated the importance of guidance and strength to encourage experimentation with disclosure. Hence, her transition from weak and vulnerable, to strong and confident, was facilitated in part by the therapists.

Many noted specific strategies and changes that they found beneficial. Pete reflected on how they have incorporated a family day out into their weekly routine, as recommended by their BFT therapists. Their willingness to continue with this demonstrates the benefit they sense from it. Indeed, for Pete, this provided a further, desirable opportunity to strengthen his relationship with the client.

Pete: That day that we go away together. That was something that came from this … we've been doing that ever since

Theme 3: Too stressed to reflect/Caregiving in a challenging system

This inductive, superordinate theme encompasses the need for caregivers to discuss the challenges of their role, despite not being asked specifically about this during interview. Caregiver's likely needed a platform to vent their frustrations and emotions, having

rarely been given the opportunity to reflect on their own caregiving experiences. Although many of these challenges do not directly relate to the process or content of BFT, they are inextricably linked. Indeed, it would be negligent to deliver therapy without considering the context in which service users are situated.

The battle for support

One of the most salient themes relates to the overwhelming pressure and responsibility on caregivers given their perceived lack of support. Caregivers felt their needs were dismissed and that services were insufficient or unacceptable. A sense of disconnect between what was being offered to caregivers and what they needed was apparent in their described frustrations.

Mary: what [client] was needing was to go into hospital. She was threatening to run out in the road and kill herself and different things like that and you're like 'oh my god' you know? There's only so much you can do…I might be a nurse and her mum, but she needs professional help

Mary's quote exemplifies her sense of helplessness with regards to keeping the client safe, despite her wealth of knowledge and expertise. She highlights the morbid risks posed but felt that this level of responsibility was beyond the remit of a caregiver. For Mary, to accept this responsibility as part of her caregiving role, would

mean to accept failure in this duty. Although Mary stressed the need for external support, she did not communicate a sense that this role should be subsumed by BFT therapists, but that it must exist adjunctively, to complement and enhance overall engagement and benefit. Similarly, Christine noted the severity of risk involved in her caring role. Despite asking for help, she felt solely responsible for the safety of the client, a responsibility that she felt should sit with trained professionals. Christine felt little appreciation for her efforts to keep the client safe, which perpetuated her frustrations with the lack of support available.

Christine: [client] told her [GP] how she was feeling, that she doesn't want to be here anymore and the doctor turns round and says to her I cannae [cannot] do nothing 'cos you're under [a different doctor]…if she kicks off…she's nowhere to go…she needed help that day, but nobody was there to help her except for me (pause) and then I got it all

This extract demonstrates Christine's frustrations and offers insight into what she wanted and needed from services. She implied a sense of being passed around by services, with no one willing to accept any responsibility for the client's safety. Again, no expectation on BFT therapists was implied. Instead the implication was that there needs to be a level of support available to take pressure off families, to

allow for better engagement in treatment, such as BFT, to improve their wellbeing. There was a strong sense from caregivers that people with intellectual disabilities are neglected within the current social and welfare system, and that too often families are left to struggle on their own. Caregivers felt as though they were 'fire fighting', which often drained them of the resources required to engage in other activity such as BFT.

Pete: You have to fight for things, when other people get handed them

Kerry: eventually they will start gradually reducing some of that support but at the moment I just keep fighting to keep that in there

Pete and Kerry's use of language reflects their sense of exhaustion from their pursuit of equality in service provision, and it illustrates their sense of battling with the welfare system. Kerry conveyed a further example of pessimism related to her expectations of others, likely resulting from experience, which assumed further withdrawal in already diminished support. Connotations of battle gives a sense of 'them against us' and demonstrates their concern about what might happen if they surrendered. This sentiment is echoed by Moira, who communicated a shared sense of hopelessness and negativity about the level of care and support available to caregivers.

Moira: the system is such that they're happy as long as there's a certain amount of functionality

Interestingly, concerns raised by caregivers predominantly related to the lack of support and resources for the client, not for their own emotional and physical needs. This reflects a recurring pattern of prioritising the client's needs above their own. However, one salient theme pertains to the lack of guidance available to caregivers. They felt that deviance from the usual life trajectory left them isolated, dismissed and feeling inferior. This was particularly prominent during the client's transition from childhood into adulthood. Caregivers felt uninformed about the resources and support available, and they felt left to manage alone when support was withdrawn.

Pete: Since he left school, there's been nothing really…In school you're mollycoddled but when [client] gets older and that, it's not the same…I don't know what's out there anymore…but there's nobody that's come out and said this is what's available to you

Sense of duty to protect

Caregivers described heightened anxieties resulting from their perception of increasing danger in the world. Concerns about client safety was raised by all caregivers throughout their interviews, though this topic was not elicited by the interview schedule. Caregivers' need to share their concerns may reflect a desire to share

the responsibility, or to elicit support, but perhaps more likely reflects the overriding nature of these worries.

The need to be on constant alert, saturated with a sense of anticipated danger, was evident throughout caregiver accounts. Many reported becoming pessimistic or suspicious of the intention of others, perhaps including BFT therapists. A sadness about this manifested during Mary's interview when she reflected on her evolved automatic cynicism.

Mary: she doesn't see the dangers like you or I … you do go through your life always being … on the alert all the time … you're very defensive

As mentioned previously, Mary's evolved identity as sole protector contributed to her increased caution and scepticism, which extended to healthcare professionals' motives. This adopted role also demanded vast amounts of energy and attention, leaving little to distribute to other tasks, or indeed therapeutic activities. Derek elaborated on this by conveying the emotional impact of anticipated danger. His experience suggested that danger was imminent, yet he felt powerless to avoid it.

Derek: there's been scores of incidents, over the last three years. So, you're basically sitting waiting for something to happen … it's frightening

Concern about the intentions and actions of others was a commonality amongst caregiver reports. Indeed, Pete presumed a shared perception of increased danger with the interviewer. He eluded to the interdependence between worry and responsibility, by offering insight into his perception of his son's limited self-awareness and capability to keep himself safe. This evoked a greater sense of duty for Pete to protect the client.

Pete: Because he [client] draws attention to himself, he's more vulnerable. And you know the kind of people going about just now

The fact that concerns were raised by caregivers during an interview about their experiences of BFT, highlights their significance. They wanted to feel understood by services and supported with the risk they faced. Kerry recalled an incident involving the client being locked in a flat with two men. This reflection triggered many difficult emotions and worries about what might happen if Kerry withdrew her protection. It also reveals Kerry's difficulties with tolerating uncertainties about the client's safety when she is not there. It is likely that sharing this information was an effort to convey the magnitude of the difficulties she experienced and to help the interviewer appreciate why continuing with BFT under this stress was too difficult for her.

Kerry: if they couldn't have got out (pause) you don't know what would happen

Kerry's pause in her quote is significant as it demonstrated a period of consideration. It is likely that a multitude of worries and fears travelled through her mind during this pause, but she chose not to vocalise them, perhaps due to fears that this would make them more real. There was a sense that Kerry also considered the potential impact that these fears would have had on her, given that she was not there to protect the client in this instance. Derek also discussed several incidents involving huge levels of risk and worry for both the client, Derek and others.

Derek: [client], he's open to abuse. He's been bullied, sexually abused, financially abused

As might be expected, a heightened sense of risk and danger led many caregivers to increase their level of control. A battle between taking control and fostering independence was identified by most caregivers, who often perceived clients to be less aware of their vulnerability. Guilt about imposed restrictions on the clients' independence, but acknowledgement of its necessity was a feature of many interviews. In the following quote, Andy highlights the internal struggle experienced between feeling reassured of the client's safety, and guilt about the costs that this has on the client.

Andy: see the day that we got told [client] can't have any children? I actually came back and went 'thank goodness' … that sounds terribly selfish

It is interesting that Andy chose to share this information with the interviewer during discussion about BFT and, despite his perception of how it may be received. The tone of this quote differed from his previous narrative, suggesting that familiarity with the interviewer and building of trust allowed Andy to share deeper thoughts, concerns, and feelings. This event was clearly significant for Andy, who felt that his relief was justified given the level of worry he had about the possibility of the client having children. Yet his self-critical statement invoked reassurance from the interviewer that would dispute it. Being a grandparent is generally perceived as a positive experience. Yet, Andy's quote demonstrates the considerations he must make as part of his duty to protect the client, and potentially to care for and protect her children too.

Other caregivers communicated a feeling of guilt regarding their increased efforts of control, which led them to conceal their efforts from the client. Some described the dichotomy of protective or controlling efforts and the desire to allow the client freedom and independence, and the difficulty of finding a balance between the two. Andy acknowledged that BFT sessions had provided a platform

for open discussions about these issues. He highlighted the benefit of having a therapist present in BFT sessions, whereby worries and concerns could be raised and managed efficiently. For Andy, the therapist acted as a neutral mediator, which allowed for difficult discussions to take place, as well as problem solving activities to address them. Andy noted that these opportunities helped him to develop a more effective way of managing issues, although conveyed a sense of guilt when these strategies involved withholding information from the client. Despite this, Andy felt that this concealment protected the client from potentially distressing information. The following quotes demonstrates this negotiation of his own sense of morality.

Andy: I'm not saying it's made me be a bit more devious, but it has made me be bit more kind of like…I just think from her [client] perspective it's better that she doesny know

The impact of the caregiver role

Although caregivers did reflect on the positive impact of their caring role, the most prominent themes concerned negative consequences of caring for someone with an intellectual disability, and the challenges these consequences impose on relationships. The impact is understandably extensive, with caregivers alluding to physical, financial, and emotional costs, as well as to consequences on

systemic relationships. Caregivers had to navigate a way of conceptualising these consequences, that allowed them to maintain loving, caring relationships with the client. It is interesting to note that the interview schedule did not directly enquire about perceived impact, which exemplifies its significance for caregivers. The breadth of impact highlights the need for support from the entire system to implement and facilitate BFT. This may extend to the client, caregivers, other family members as well as the therapists and indeed those further removed, such as employers and other healthcare professionals. Caregiver's need to discuss these issues dominated their need to reflect on BFT, as illustrated in the following extract.

Interviewer: So is there anything else that you can think of about...the BFT bit that you think is important to say…

Kerry: no I can't cos…I've kinda been under a lot of stress…a lot of stress lately…with my own job, my own health

Kerry exemplifies the disabling effect that stress can incur on one's ability to reflect. The difficulty she exhibited in recounting her experiences of BFT denotes the significance of other stresses in her life i.e. her employment and health. Like many caregivers, Kerry attributed deterioration in her physical health to the stress involved in her caring role. Her frustrations about these negative

consequences, and perhaps a painful fear related to her own role in their development, led to a need to blame someone or something for her circumstances. Kerry, like all caregivers, oscillated between directing this blame towards the client, and empathising with the client's difficulties and then blaming other factors.

Moira also discussed the physical impact of her caregiving role and worried about the escalating effect that deterioration in her health might have.

Moira: you feel as though if we don't get this in place, the stress of it all could shorten our lives … my husband's health was kinda suffering a bit with that. He had a (pause) wee heart problem

Moira emphasised the potential effect of enduring stress, but minimised the severity of its existing impact. This enabled her to accept, and cope with the negative consequences suffered by her family as a result of her caregiving duties. Acknowledging the potential consequences of continued stress, served to motivate Moira to change the situation to prevent any further impact, rather than to place blame with any one individual. However, abating the severity of her husband's health problem may serve to mediate any implication of blame on the client. Reframing the potential costs of her caregiving role, relieved potential cognitive dissonance arising

from providing love, care, and compassion for the client, whilst also recognising the negative impact her caregiving role may produce.

In the following extract, Mary also demonstrates her struggle with recognising that difficulties with her health and employment were largely attributable to the client, but also wanting to maintain empathy and deflect blame away from them. Mary's benevolence also endorses a loving and caring relationship with the client, despite presenting difficulties.

Mary: I've had a few sickness absences and I don't like apportioning the blame to [client] but it's because of the situation at the time

Kerry described her experience of continuing to work whilst struggling with her caring responsibilities. She encapsulates the difficulty of focusing on routine activities whilst under immense pressure and stress. Her caregiving role was all consuming and left her feeling incapable of committing to other tasks, including BFT.

Kerry: it was absolutely horrible … I couldn't concentrate on my job … I was crying all the time … I would give up jobs to come back and look after [client]

The stress of her caregiving responsibilities not only affected Kerry's ability to maintain employment, she also felt that familial relationships were affected. Although some caregivers believed that the challenges of their caregiving role strengthened their bond with

their partner, Kerry attributed the difficulties in her marital relationship to the stress of the caregiving role.

Kerry: it put a strain on our relationship [with her husband] for a long time

Kerry also conveyed a sense of guilt regarding unpleasant situations that the client's siblings experienced as a result of the client's behaviour. This contributed to their absence in BFT sessions. It is therefore interesting to consider how Kerry's conceptualisation of the client's impact on familial relationships may have manifested during BFT sessions, had they all participated. Indeed, seven of the interviewed caregivers also cared for siblings of the client, and all highlighted the difficulties in managing their time, energy and attention between them. A sense of guilt about devoting more time to the client was a clear theme amongst these caregivers.

Mary: she [client] gets all the time spent on her (pause) probably at the detriment of my other daughter

Moira felt that strain was placed on her daughters' relationship due to the observable consequences the caregiving role was having on her and her husband. She felt it was important that siblings' needs and perspectives were heard and valued, and suggested incorporating an additional session of BFT, devoted to client siblings to address this.

Frequent reference to the sacrifices made by caregivers in order to care for the client was evident. Many described a change of identity following the birth of the client, to one that revolves around their caregiving duties, rather than the caregiver as an individual. Mary demonstrated a complete dissociation with her pre-caregiving self by describing her lack of personal time as life-long. She paused to consider her choice of words, to prevent any sense of regret or ungratefulness regarding her caregiving role. Yet, a sense of mourning for her expected, or 'normal' life was evident.

Mary: I'm 64 and [husband] is 67 and all our lifetime we've not really had any like (pause) 'me time' as I would call it?

As mentioned previously, BFT itself requires a degree of sacrifice in terms of the time, commitment, and arguably, privacy required. In other words, to implement BFT appropriately, a trade-off with other commitments or activities must occur. Andy conveys a feeling of sympathy and admiration towards his wife for the level of sacrifice she made to support the client with BFT. He emitted an air of desperation in his perception of his wife's devotion and desire to help the client in his repetition of the word 'anything'. This indicates that Andy also feels his wife's efforts may exceed the expectations and remit of the caregiving role.

Andy: my wife was giving up…big elements of what was her only day off during the week…she would do anything (pause) anything

Caregivers often felt disregarded or criticised despite their sacrifices and efforts. They wanted what they perceived to be a normal, loving relationship with the client, but often felt hurt and confused by their behaviour, which impacted on their ability to interact in the loving, compassionate way they desired.

Diane: See when that red mist takes over? All hell breaks loose … once he's [client] got over that … he'll say Mum I'm sorry, I didn't mean to do that. Can I have a cuddle?

*Christine: she can be very hurtful … she'll say "I f*cking hate you" like that and then she'll just turn round and say mum look, I'm sorry that I said that*

Diane and Christine's account of the contradictory verbal and physical communication of the client reflects the contrasting feelings they are left with as a result. Their process of coping required depersonalisation of their comments and behaviours, which was supported by the client's apology. Both described deep love for the client, but an unpleasant disdain towards them, which had to be processed and framed within the context of their intellectual disability to be accepted and tolerated. BFT gave them and the client an opportunity to express these feelings in a supported and

structured manner. They were encouraged to shared difficult feelings that they would have otherwise suppressed, in order to resolve issues where possible.

BFT also offered an opportunity to spend time together as a family, which Pete valued. Pete also described difficulties in maintaining a close, loving relationship with the client, due to the characteristics and symptoms associated with the client's intellectual disability. Pete described being rejected by the client as 'heart breaking', signifying the continued emotional impact of this issue.

Pete: it's kind of heart breaking at times that he doesn't want to be associated…I always think that father and son should have some sort of bonding…I want to spend time with him- I want to do this. But you just feel shunned all the time

Pete's prediction of rejection by the client and the emotional impact of this, reduced his motivation. His quote bears an element of hopelessness about the situation improving, perhaps due to difficulties in him processing or accepting this reality, but also a prevailing desire to develop and maintain a loving relationship with the client.

"Who's going to fight for him?": The future beyond my existence

Although hope permeated many interviews, worries about the future tended to overshadow this positivity. Overwhelming concern about the clients' future was a significant contributor to caregivers' daily stress. Although worry and disdain was a feature of discussion around current support provision, a different sensation was evident during consideration of the client's future risk, particularly when considering increased likelihood of physical deterioration associated with ageing. Many caregivers referred to life beyond their existence and the challenges that may emerge without their input. Thoughts about mortality were raised by most caregivers with regards to their own worries, as well as worries articulated by the client.

Diane: He [client] says Mum, what's going to happen to me when you and my Dad aren't here?

Diane framed concerns about future provisions as a worry belonging to the client. However, her delivery of this statement demonstrated a shared concern. Interestingly, Andy also raised this concern as belonging to the client. Their strategy for coping with this morbid concern allows them to disengage with it, whilst recognising its existence. To verbalise these worries as their own, may be too difficult for caregivers, as it forces them to engage with this painful reality. Andy's interpretation of the client's statement infers a worry about what the client would do after his death, which is likely to be

influenced by the client's previous suicidality. It also demonstrates his experience of pressure and responsibility created by her dependence on him, as though for Andy, the continuation of his support truly is a matter of life and death.

Andy: she [client] said to me…I hope we all die at the one time…that's what she says…I hope we all die at the same time…I don't want to be here if you're not here basically.

Caregivers with other children, communicated huge concern and often guilt, relating to the expectations and pressures on these siblings after their death. Experienced difficulties in sourcing sufficient current support and the assumed added complexities likely to surface with age, were of particular concern. The expectation that siblings may be involved in supporting clients is interesting, given both siblings themselves, and caregivers' reluctance for them to be involved in BFT.

Diane: When your Dad and I are not here…we're not expecting you [sibling] to look after your brother [client], I says, but he is your brother…I'm wanting to make sure that you're gonna phone him and you chap his door. I don't care if he slams the door in your face

Here Diane presents a concern that the client's sibling will not adopt the level of responsibility required to care for the client after her death. Her slightly contradictory language in first saying she does not

expect the client's sibling to take on caregiving duties, but then continuing to describe what that duty would involve, reflects her internal battle between needing reassurance of the client's safety in her absence, yet not wanting to assign any burden to her other son. Diane anticipates challenges this transfer of responsibility will incur and conveys a worry that this might dissuade the client's sibling from assuming this duty. She intimates a fear that the client will be neglected and alone without her there to support him.

Derek's concerns related more to keeping the client safe from potential outcomes arising from his intellectual disability. Throughout his interview, Derek spoke of the client's limited recognition of the consequences of his own behaviour. Derek felt he had to intervene on many occasions to protect the client from harm and that the level of harm was continually intensifying. Derek's perception of the client's lack of insight and vulnerability led to an increased sense of duty and worry. His perception of the level of risk posed by these vulnerabilities was also escalating with time. Indeed, Derek conflates maturity with increased risk in the following extract.

Derek: the difference now between eh (pause) when he was hooking in with people when he first came here and he was 16, when he's hooking in with them now … it's a lot more mature now. (Pause) he's hunting (pause) hunting now specifically for sex

Increasing worry also led to grave concern about dangers posed by the world around the client as well as the people in it. Given caregivers sensed duty of protection and their mounting perception of dangers, considering the client's future without their protection was difficult. In fact, Mary preferred to avoid this fate, by hoping she would outlive the client.

Mary: I hope I don't die before her because I don't want to leave her on this earth, because (pause) people are cruel … the dangers are more prevalent

Although Mary's quote assumed a desire that the client dies first, her language conveys a kindness and compassion for the client. She paused to consider how to justify this hope, then confidently used a bold, generalising statement that epitomises Mary's perception of the world. Mary's pessimism of the world, as demonstrated by her quote, extends to all outside of her family, which transmits a feeling of grandeur, that assumed they are the only ones that can provide adequate support. Indeed, many caregivers believed that no one else would be eligible to subsume this role. This enormous sense of responsibility augmented worries about provisions after their death.

Pete: As he gets older and we're not here to fight for him, who's going to fight for him?

Pete echoes the sentiment from many caregivers regarding the transfer of the immense caregiver responsibility following their death. His question indicates that a plan for future provision had not been confirmed, despite their huge concerns. Interestingly, none of the caregivers referred to any plan for future provision, possibly highlighting the denial, or difficulty, caregivers experience when considering this reality.

Discussion

This qualitative study explored caregivers' experiences of being a BFT participant. Understandably, caregivers' views were variable, however several key themes emerged. These themes offer valuable insight into the unique perspectives of these caregivers and their journey to, and through the BFT programme, which can help to inform service planning, provision and delivery. Each theme will now be discussed in relation to existing literature in this field.

Journey of accepting and engaging in BFT

Given the abundance of research that highlights the difficulties in successfully implementing behavioural interventions in clinical settings (Hutchison et al, 2017; Michie et al, 2007; Campbell, 2000; Fadden, 1997; Kavanagh et al, 1993), it is necessary to understand contributory factors in enabling or hindering engagement.

Organisational barriers such as staffing, resources, and resistance, are well documented (Absalom-Hornby et Al 2011; Bailey et al, 2003; Smith & Vellman, 2002), but less is known about the service users' perspective, or caregivers' specifically.

The need for service users to trust that their therapists understood their needs and could help meet them, are familiar features within engagement literature (e.g. Odell & Quinn, 1998) yet this study presents a picture of caregivers' complex and unmet needs. Caregivers described constructing their own barriers to defend against judgement and to cope with their fears of being criticised for their caregiving abilities. They needed to trust their therapist in order to feel safe in deconstructing this defensive coping strategy. They also needed to trust that this challenge would be worthwhile and that their therapist had the skills to support them through it. Finally, they needed to trust that they would not be criticised and judged for revealing previously hidden thoughts, feelings and worries. This was particularly challenging for caregivers who held negative assumptions and scepticism about the perspectives and motives of others.

Caregivers in this study noted the benefits of validation and empathy from their therapists, which is reflected in engagement literature (e.g. Beeber et al 2007). Some suggest that meeting others who have shared similar experiences is helpful (Rose, 1998; Singer, 1999).

Certainly, this was suggested by one caregiver in this study. However, caregivers' descriptions of their battles with seeking and accepting support in this study have unique characteristics. Hesitation to express the frustration and desperation related to their caring role, coupled with a strong, sometimes rigid expectation that they should manage just as they perceive others to, perhaps necessitates increased validation and empathy from therapists. Trust, gentle encouragement and support from therapists can help to facilitate progression to a position of readiness and preparation for change (The transtheoretical model; Prochaska & DiClemente, 1983).

The timing of intervention appears crucial in caregivers' ability to engage optimally in BFT. For some, a feeling of complete helplessness and desperation was necessary for support to be perceived as acceptable. Certainly, research indicates that families are more likely to engage in therapy following a crisis (McCreadie, 1991). Conversely, there was a sense from caregivers that a degree of stability was necessary to allow the capacity for engagement. Indeed, many believe that immediate risk must be managed before effective therapeutic change can be contemplated, particularly when working with suicidal clients. However, one can argue that moments of crisis represent the most crucial period for intervention implementation. As highlighted in this study, stability is often atypical, with frequent legal concerns, housing difficulties, injury and suicidality featuring in

their lives. For these individuals, it may not be reasonable to withdraw intervention each time risk arises, or to deny them the opportunity to engage at all.

Some caregivers were more accepting of their need to adapt their own behaviour to support overall positive change than others. As documented in psychosis literature, many reported a sense of benefit and empowerment from the strategies they had learned from BFT, that promoted improvements in communication for the entire system (Budd & Hughes, 1997). However, some caregivers attributed the bulk of systemic problems to the client alone, thereby obstructing or revoking any responsibility or motivation to address their own roles and behaviour. Some caregivers in this study posited that engagement from all members of the system promoted unity and connectedness, though united engagement also helps to distribute caregiver burden (Murphy et al, 2007). Caregivers in this study who were more positive about their experiences of BFT, highlighted the need for shared ownership over the system's progress. Absence of this shared responsibility may help to explain some of the difficulties in engaging families in BFT, as highlighted in previous research (Anderson et al, not published).

Although many caregivers acknowledged therapeutic benefits of BFT for all involved participants, there was a consensus that intervention

was predominantly designed for, and necessarily tailored to clients' needs and capacity. Although adaption of materials was perceived positively, some caregivers' perspective on the client's ability to engage and effect change from therapy more generally, reflected the historic assumption that people with intellectual disabilities cannot benefit from therapy. Indeed, as is documented in previous literature (Pharoah et al, 1999), many caregivers inferred that therapeutic change was minimal and evident in their behaviour only.

Caregivers in this study reflected specifically on the slow pace of therapy, a finding supported in previous research (Goldberg et al, 1995). Yet research indicates that a slow pace is required not only to synchronise with the clients' processing speed, but to allow the therapist time to develop a deeper understanding of family dynamics. A slow pace also allows time for the family to process multiple experiences of loss that may resurface throughout life, such as the loss of 'the perfect child' (Oswin, 1991; Olsson & Hwang, 2001), expected grandchildren, or a peaceful retirement (Goldberg et al, 1995). Despite this rationale, caregivers in this study communicated continued grief and pain related to their experiences of loss. This may indicate inadequate processing, particularly of the loss of the desired relationship with the client and of future hopes and expectations for them.

Existing research on caregiver experiences of caring for an individual with an intellectual disability, indicates that caregivers can struggle with accepting the diagnoses of their loved ones (Hastings & Beck, 2004). One of the most thought-provoking themes in this study, was the caregiver's perception of the client's ability to accept their intellectual disability diagnosis. It is interesting to explore the origins of this refutation, and how it might impact on the whole family's willingness to engage in intervention. Indeed, caregiver knowledge and insight may influence the insight of people with long term health difficulties (Brent et al, 2010). It is possible that caregiver's initial difficulties with acceptance has led to the client's rejection of their diagnosis, both of which may derive from negative experiences associated with intellectual disability, and the socially constructed stigma related to it. It would not be surprising if, given their desired dissociation from any affiliations with intellectual disability, clients were reluctant to engage in BFT, which would impact on caregiver's ability to engage. Certainly, those who do not willingly seek therapy are rarely invested in making changes (Kazdin et al, 1990).

Sense of Progression and Change

Part of the difficulty experienced by caregivers related to the permanent nature of intellectual disability. A sense of denial of this permanence pervaded some caregiver accounts, which led to

frustration about their incapability to improve their circumstances. This in turn, contributed to increased feelings of hopelessness. The psychoeducational element of BFT aided caregivers' understanding of intellectual disability and the potential extent of its impact on the client. Caregivers in this group who could acknowledge and accept the pervasiveness of certain client characteristics, and attribute them to their intellectual disability rather than the individual as a person, appeared more capable of redirecting their energy towards factors that could be changed. Increased understanding of intellectual disability led caregivers to be more realistic about the level of change that could be achieved and an increased tolerance of difficult behaviours. This also supported the maintenance of empathy and love felt towards the client. Many caregivers also reflected on how BFT supported them to reframe negative assumptions, and to consciously consider the positive aspects of their situation.

Many caregivers in this study reflected on how they felt their caregiving competencies were enhanced by the skills they have developed in sessions. This confidence empowered them to experiment with different strategies to manage stressful situations, enhance problem-solving efforts, and generally improve system relationships. This reflects existing literature on family interventions (Bird et al, 2010) and is significant given that caregiving satisfaction levels, positive relationships, and increased internal locus of control

can mediate experiences of stress, depression and anxiety (Yoong & Koritsas, 2012; Hill & Rose, 2009). Some caregivers attributed improvements in relationships to developments in their communication skills. Advances in communication allowed the caregiver and client to share their internal world and to foster deeper understanding of each other's perspectives. This also encouraged a dialogue in which problems could be shared and resolved collaboratively. For some, positive outcomes of BFT led to increased client independence, which is echoed by literature on other family programmes (Daley et al, 2015).

Caregiving in a challenging system

Although inductive themes lightly permeated interviews, this final theme focusses on the significant issues raised by caregivers that provide a rich context from which their experiences of BFT can be better understood.

Caregivers wanted clearer information about the support available, and earlier intervention and support, not just during crisis, as highlighted by the Carer's Strategy for Scotland 2010-2015. However, it is clear from this study and other research (McGill et al, 2006a; Lafferty et al, 2016) that this appeal has not been adequately addressed. Caregivers in this study reflected on their sense of disconnect with what was needed, and what services provided. This

is important, as better access to services contributes to improved caregiver wellbeing and greater confidence in their perceived ability to cope (Holl & Morano, 2014).

Caregivers articulated a feeling of pressure to care for the client without support, and many expressed dissatisfactions with the support that was provided, which is reflected throughout relevant literature (Findlay et al, 2015; McGill et al, 2006b; Robertson et al, 1996; Ruef et al, 1999). This highlights the confusion, or ambiguity relating to where the responsibility for individuals with intellectual disability should lie. It also implies the difficulties caregivers face in seeking support from professionals to share this responsibility. This is likely to feed into their pessimism about the world and others, and further fuel their frustrations with inequalities experienced by this group.

Caregivers reported increased stress in relation to the client's transition into adulthood. Existing literature corroborates an increased difficulty in accessing services during this period (Blomquist, 2006), which also reflects previous literature on caregiver experiences (Pilnick, Murphy & Almack, 2011). This theme may demonstrate an anti-austerity message, reflecting diminishing funding provisions available at present. Indeed, there has been a marked increase in demand on services like respite support in recent

years, but service provisions have diminished (McConkey et al, 2010). This theme may also signify a gap in service provision, reflecting a dismissal or misunderstanding of the needs of individuals with intellectual disabilities and the systems that support them.

Caregivers all referred to increased cynicism about the world and the dangers clients are exposed to. This led to conflicting feelings regarding the dual imperatives of control and independence, a difficulty shared by families in existing literature (Almack et al, 2009' Chadwick et al, 2013). With more people with intellectual disabilities now living at home, complex and emotional decisions regarding independence are left to their caregivers, who may be left feeling helpless, guilty, and distressed about their choices. Caregivers wanted support and guidance to manage these moral and emotional dilemmas and to make the necessary adjustments to their expanding remit as a caregiver, which many felt was partly provided through BFT. This supports the 'adaption hypothesis' (Seltzer & Krauss 1989; Grant 1993), whereby caregivers acclimatise to evolving conditions of their role, despite diminished or non-existent support. However, caregivers in this study indicate that BFT helps to facilitate this adaption.

Caregivers' conceptualisations of the world also led to intensified worry and anticipation of danger. They described feeling as though a

perpetual state of alertness was required to maintain client safety and to effectively respond to unpredictability. This is reported elsewhere by caregivers of people with autism (Butrimaviciute & Grieve, 2014), who described the exhaustion of intense physical and mental engagement to ensure the safety of clients and others. This intense engagement and exhaustion may account for caregiver's difficulty in this study, to engage in BFT and, indeed, reflect on their experiences of it.

As previously discussed, caregivers for individuals with intellectual disabilities experience greater stress (Emerson, 2003), but the impact of their caregiver role is far-reaching. Research on caregivers experiences of caring for an individual with an intellectual disability, indicates that many felt they had lost touch with the outside world, they felt disempowered, their social networks diminished, their physical health deteriorated more rapidly, and they felt that increased strain was placed on family relationships (Fidell, 2000). All these experiences were shared by the caregivers in this study and were significant enough to discuss during their interview concerning BFT. This demonstrates the importance of satisfying lower-level needs to allow for higher level processing and reflection to occur (Maslow, 1943). It may also explain why engaging families under this level of stress is so challenging. Adequate social support would help to mediate the strain experienced by caregivers (Tsai et al, 2009; Hill

& Rose, 2009), but this was difficult for these caregivers to source. The absence of adequate support left caregivers to struggle alone. Caregivers described the challenges this posed to maintaining employment, which then impacted their financial stability (Parish et al, 2004). This is significant, as it is suggested that caregiver psychological distress may be mediated by socioeconomic factors (Emerson, Robertson & Wood, 2004).

The impact of their caregiving role on familial relationships was significant to many caregivers in this study. Some attributed difficulties to the stress and burden of their role, as well as the energy and attention it demands, often leaving little for other family members. Concern over the impact their caregiving role had on siblings of clients, was raised by many caregivers. Furthermore, research indicates that the stigma associated with intellectual disability may also affect family members (Birenbaum, 1970/1992). This 'courtesy stigma' can lead to siblings being teased (Larson & Corrigan, 2008), which is likely to cause difficulties in their relationship with the client. Just one client's sibling in this study engaged in BFT, which may support this hypothesis.

Finally, tolerating uncertainty, and fear of the future for adults with intellectual disability has been documented in previous research (Eley et al, 2009; Price et al, 2017). Indeed, many caregivers in this

study chose not to think about the future due to worry and uncertainty about the client's ability to care for themselves, their health, and levels of service provision. Some chose to focus on positive prospects for the client, despite having no assurance that their hopes would materialise. Furthermore, many caregivers continue to look after clients well into the 70s. Yet, as demonstrated in this study, very few caregivers develop concrete future care plans. This means that support packages are frequently designed during crisis, with little planning and preparation (Prosser, 1997). Support in managing their fears and expectations of the future, was provided for some through BFT, in that it improved communication and problem-solving skills, which led to greater confidence on the part of the caregiver and client. Indeed, many caregivers acknowledged advances in the client's level of independence, which instilled hope for the future.

Limitations and future research

This study provides valuable insight into caregivers' experiences of being a BFT participant, though several limitations are acknowledged. Firstly, selection bias may have occurred in the recruitment phase, as Team Leads were responsible for inviting candidates to participate in the first instance. It is possible that caregivers deemed to possess a

more positive view of their experience of BFT, were preferred over those assumed to hold negative views. It is also possible that those willing to participate in the study, held strong positive or negative views, which may again affect the findings.

Although every effort was made to ensure that participants were aware that their consent to take part would not affect their family's care, it is possible that this was a concern for some, who may be more inclined to report positive reflections on their service. Similarly, those interviewed as a couple may have been constricted in their freedom of speech. Undesirable thoughts and opinions may have been withheld due to concerns of the other's response.

This study considers the experiences of 7 parents and 1 foster carer, who had engaged in a minimum of 5 sessions of BFT. This sample is relatively small therefore it is not possible to generalise the results to wider populations. It is possible that significant experiences have been missed, therefore future studies could include a larger sample to capture a wider variety of views and experiences. The voices of those who dropped out before session 5 and those who were never known to services in the first instance are neglected. Future research could explore the views of caregivers who dropped out of therapy before this point, or who refused to take part at all, to better understand their views. Additionally, cultural diversity is limited with

all caregivers being White and British. It is likely that other cultures experience different norms and treatment. Further research could involve caregivers from broad backgrounds to reduce this limitation. Furthermore, this study does not comment on the level of client intellectual disability. Some research indicates that this may be a predictor of carer wellbeing (Abbeduto et al, 2004), therefore may also contribute to their experiences of receiving, engaging in, and benefiting from BFT.

Caregiver experiences were considered, partly due to the level of involvement they assume in the client's life, and the importance of their involvement in BFT when evaluating outcomes. This study however, forms part of a larger research project investigating the utility of BFT in intellectual disability services more generally. Staff experiences have been investigated, yet research has not yet explored siblings or indeed client's perspectives. Future research directly investigating client experiences of BFT would aptly complement this study, particularly as increasing evidence indicates that despite likely difficulties with communication, people with intellectual disabilities can provide informative, qualitative data (MacMahon et al, 2015). Indeed, an increasing amount of research is investigating their experiences (Woolfall, 2018).

A larger, quantitative study using validated outcome measures may help to objectively measure the perceived changes reported by caregivers in this study. Outcomes such as the Caregiver Strain Questionnare (CGSQ; Brannan et al, 1997) have been successful in measuring changes following BFT in a case study example (Marshall & Ferris, 2012), therefore may provide a good starting point to explore significant change at cohort level.

Finally, it is recognised in IPA that the researcher brings their own biases to both their interactions with participants and to their interpretations of participant experiences. Caution must be taken when making conclusions about these interpretations, particularly when considering the bias that may arise when working with a marginalised population.

Clinical Implications and Recommendations

This study sought to explore caregivers' experiences of being a BFT participant. It was hoped that a deeper insight into their experiences might illuminate the areas of BFT delivery that may be improved, as well as how we might endeavour to achieve this. The clinical implications of this study's results, along with some possible recommendations, will now be discussed.

Selling BFT

- Alignment of therapist and participant expectations of BFT could be considered prior to initiation. Potential difficulties with logistical, emotional, and motivational obstacles should be addressed. A pre-engagement session may provide this opportunity. This would also help to alleviate anxieties about engaging, enhance understanding of the process of BFT, and allow for the trusting, therapeutic relationship to develop.
- Some caregivers indicated that crisis was a necessary prerequisite to engagement, while others considered that risk must be managed sufficiently first. It is interesting to consider whether BFT could be offered to all families who feel that although they are managing, things could be improved. This may alleviate any guilt or shame regarding accepting therapy.
- Many mentioned having worked with their therapist for a long period of time before starting BFT, suggesting it is often used as a last resort, following failed attempts utilising different approaches. It may be more appropriate for families to attend BFT as an optional precursor to individual therapy to ensure that the appropriate support is in place to promote benefit.
- The communal focus of treatment should be adequately conveyed from the outset to engage all members, and alleviate blame apportioned to the client alone. It is possible

that families who benefit most from BFT are those who all accept some responsibility for promoting and effecting change.

Therapist Considerations

- It is essential to consider the emotional turmoil that caregivers are likely to have suffered both prior to referral, and at the point of assessment, to ensure therapist sensitivity, understanding, and empathy. Therapists must also consider the current impact of stress in relation to families' ability to commit and engage.
- Caregivers' prior experiences of support may taint their expectations and trust in any future intervention. Similarly, caregivers described increased pessimism about the world and the intentions of others, which may extend to BFT therapists. Addressing this potential concern may help to reassure caregivers and reduce its impact.
- Simply by listening intently to the client, the therapist can encourage other members of the family to follow suit (Fidell, 1958). However, therapists must reach a balance between empowering and advocating for the client, whilst appreciating that clients may have been used as a scapegoat for familial difficulties for many years. Acts of empowerment may

alienate other family members, disrupting the therapeutic alliance.

- It is particularly important to foster positive, healthy relationships between family members where siblings are present, given the increased likelihood of resentment and other negative feelings between siblings (Shivers & Kozimor, 2017). It is becoming increasingly important that adequate support is provided to siblings of people with intellectual disabilities, given the likelihood that they will subsume a compound caregiving role at some point in the future, caring for their ageing parents and their sibling simultaneously (Tebes & Irish, 2000).
- Given the number of complex factors to consider when working with this population, advanced BFT training opportunities for therapists may help to enhance the appropriate skills and increase their confidence in working effectively with this group.
- Carers frequently described feeling helpless, with little sense of agency. Increased client and family involvement in intervention delivery and care planning may help to empower families.

System Changes

- Progression towards parity of esteem is necessary for families to have equal access to resources and support for their families.
- Transition from child services requires a future planning programme that explores the clients' expectations and future goals, the level of support required, and the sources of support available. This will ensure a smoother transition to adult services. Improvements in the level of advice in sourcing support is required at this time, as well as throughout adulthood. This may also involve education and support around potential dangers in their current environment.
- A long-term life plan should be considered as standard when transitioning into adult services. This should reflect likely deterioration, frequency of check-ins for family and client, a plan for when caregivers cannot cope, and details of what each member would like to happen in the future. This will prevent ad hoc crisis management, resulting in poorer outcomes.
- There needs to be an out-of-hours service, and appropriate back-ups when the clients' doctor is unavailable. Crisis does not only occur Monday to Friday between 9 and 5.

By exploring experiences of being a BFT participant, this study consequentially taps into the lives and experiences of caregivers of

an individual with an intellectual disability more generally. Caregivers narratives indicate that they need to feel listened to. They need to feel valued and validated. They need to be managed with care and compassion. They need support to manage their stress levels and to reduce the clients' distress. They need to know how to manage risk effectively during crisis and to have sufficient support when they are unable to manage alone. They need to feel reassured that the client is safe from harm, and well protected. They want the client to live as independently as possible. They need support for their families, who may also assume caring responsibilities for the client. They need the opportunity for respite. While it is not expected that BFT can meet all of these needs, it is possible that it is the best we can offer in the current climate and scope of service provision. However, caregivers report that BFT does begin to address some of these needs, which is a positive start.

Conclusions

When asked about their experiences of BFT, caregivers discussed the necessary conditions to promote engagement and acceptance of therapy and their sense of progression and change following the intervention. However, caregivers also unanimously discussed the difficulties and stresses that form a large part of their caregiving role.

The impact of this stress is profound and extensive, and significantly affected their ability to engage with BFT, and indeed to function more generally. This inductive theme represents the overarching significance of this stress for caregivers.

Many people with intellectual disabilities have been marginalised, living restricted lives permeated with prejudice and stigma (Ali et al, 2012). BFT aims to be non-judgemental and to encourage consideration of how others live, think, and feel, before making assumptions. For some caregivers, BFT acted as a vehicle for adjustment, who felt that a better understanding of the client's disability and needs helped them to adjust their expectations of both themselves and the client. The purpose of BFT is clearly not to 'cure' the client of their disability, but to improve overall wellbeing of the entire family. BFT aims to achieve this by reducing stress within the family, improving communication between family members, improving their problem-solving skills, and by facilitating the identification and pursuit of individual goals. Most caregivers in this study noted improvements in these areas. They also felt that BFT provided a safe environment for difficult discussions to occur, an environment which could be replicated by the system out with sessions. The template for discussion provided by BFT promoted listening, understanding and problem-solving, which helped to

effectively address issues of concern from any member of the system.

Unfortunately, BFT cannot change the difficulties faced by clients in the past, and cannot fully eliminate worries about future support provision, but caregivers note that it can empower them in the present, and give them skills and knowledge that they can utilise throughout their lives.

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Appendices

Appendix 1: CASP Checklist for Qualitative Research and RCTS

Qualitative Research

Section A: Are the results valid?		
1. Was there a clear statement of the aims of the research?	Yes	
	Can't Tell	
	No	
Comments:		
2. Is a qualitative methodology appropriate?	Yes	
	Can't Tell	
	No	
Comments:		
Is it worth continuing?		
3. Was the research design appropriate to address the aims of the research?	Yes	
	Can't Tell	
	No	
Comments:		
4. Was the recruitment strategy appropriate to	Yes	

the aims of the research?		
	Can't Tell	
	No	
Comments:		
5. Was the data collected in a way that addressed the research issue?	Yes	
	Can't Tell	
	No	
Comments:		
6. Has the relationship between researcher and participants been adequately considered?	Yes	
	Can't Tell	
	No	
Comments:		
Section B: What are the results?		
7. Have ethical issues been taken into consideration?	Yes	
	Can't Tell	
	No	
Comments:		
8. Was the data analysis sufficiently rigorous?	Yes	

	Can't Tell	
	No	
Comments:		
9. Is there a clear statement of findings?	Yes	
	Can't Tell	
	No	
Comments:		
Section C: Will the results help locally?		
10. How valuable is the research?		
Comments:		

Randomised Control Trials

Section A: Are the results of the trial valid?		
1. Did the trial address a clearly focussed issue?	Yes	
	Can't Tell	
	No	
Comments:		
2. Was the assignment of patients to treatments randomised?	Yes	
	Can't Tell	
	No	

Comments:		
3. Were all of the patients who entered the trial properly accounted for at its conclusion?	Yes	
	Can't Tell	
	No	
Comments:		
Is it worth continuing?		
4. Were patients, health workers and study personnel 'blind' to treatment?	Yes	
	Can't Tell	
	No	
Comments:		
5. Were the groups similar at the start of the trial?	Yes	
	Can't Tell	
	No	
Comments:		
6. Aside from the experimental intervention, were the groups treated equally?	Yes	
	Can't Tell	
	No	

Comments:		
Section B: What are the results?		
7. How large was the treatment effect?		
Comments:		
8. How precise was the estimate of the treatment effect?		
Comments:		
Section C: Will the results help locally?		
9. Can the results be applied to the local population, or in your context?	Yes	
	Can't Tell	
	No	
Comments:		
10. Were all clinically important outcomes considered?	Yes	
	Can't Tell	
	No	
Comments:		
11. Are the benefits worth the costs?	Yes	
	Can't Tell	
	No	
Comments:		

Appendix 2: SIGN checklist for Controlled Trials

Methodology Checklist 2: Controlled Trials

Study identification (*Include author, title, year of publication, journal title, pages*)

Guideline topic:	Key Question No:	Reviewer:

Before completing this checklist, consider:

1. Is the paper a **randomised controlled trial** or a **controlled clinical trial**? If in doubt, check the study design algorithm available from SIGN and make sure you have the correct checklist. If it is a **controlled clinical trial** questions 1.2, 1.3, and 1.4 are not relevant, and the study cannot be rated higher than 1+
2. Is the paper relevant to key question? Analyse using PICO (Patient or Population Intervention Comparison Outcome). IF NO REJECT (give reason below). IF YES complete the checklist.

Reason for rejection: 1. Paper not relevant to key question ☐ 2. Other reason ☐ (please specify):

SECTION 1: INTERNAL VALIDITY

In a well conducted RCT study…		*Does this study do it?*	
1.1	The study addresses an appropriate and clearly focused question.	Yes ☐ Can't say ☐	No ☐
1.2	The assignment of subjects to treatment groups is randomised.	Yes ☐ Can't say ☐	No ☐
1.3	An adequate concealment method is used.	Yes ☐ Can't say ☐	No ☐
1.4	The design keeps subjects and investigators 'blind' about treatment allocation.	Yes ☐ Can't say ☐	No ☐
1.5	The treatment and control groups are similar at the start of the trial.	Yes ☐ Can't say ☐	No ☐
1.6	The only difference between groups is the treatment under investigation.	Yes ☐ Can't say ☐	No ☐
1.7	All relevant outcomes are measured in a standard, valid and reliable way.	Yes ☐ Can't say ☐	No ☐
1.8	What percentage of the individuals or clusters recruited into each treatment arm of the study dropped out before the study was completed?		
1.9	All the subjects are analysed in the groups to which they were randomly allocated (often referred to as intention to treat analysis).	Yes ☐ Can't say ☐	No ☐ Does not apply ☐
1.10	Where the study is carried out at more than one site, results are comparable for all sites.	Yes ☐ Can't say ☐	No ☐ Does not apply ☐

SECTION 2: OVERALL ASSESSMENT OF THE STUDY

SECTION 2: OVERALL ASSESSMENT OF THE STUDY		
2.1	How well was the study done to minimise bias? *Code as follows:*	High quality (++)☐ Acceptable (+)☐ Low quality (-)☐ Unacceptable – reject 0 ☐
2.2	Taking into account clinical considerations, your evaluation of the methodology used, and the statistical power of the study, are you certain that the overall effect is due to the study intervention?	
2.3	Are the results of this study directly applicable to the patient group targeted by this guideline?	
2.4	**Notes.** Summarise the authors' conclusions. Add any comments on your own assessment of the study, and the extent to which it answers your question and mention any areas of uncertainty raised above.	

Appendix 3: Quality Rating Scoring Criteria

FACTOR	WEIGHTING		
	2	1	0
Research Question	Defined including the specific outcomes explored	Defined but with no specific outcomes noted	No clear question or aims
Methodology/Design	Methodology and design are appropriate with sufficient rationale behind choice	Methodology and design are appropriate, no explanation for choice	Methodology and design are inappropriate, or unclear
Inclusion/Exclusion criteria	Criteria documented explicitly	Partially reported, e.g. just one criterion explicit	Unclear or not reported
Recruitment/sampling Methods	Sampling from more than one source	Volunteer sample or sample from one type of source only e.g. similar clinics in one area or similar schools in one area	Unclear or not reported
Randomisation	Evidence of randomisation	No randomisation, but rationale for this e.g. only one intervention group	No randomisation
Control Group	Control group similar to experimental group at baseline, e.g. age-matched, or variables such as socio-economic status	Control group present but no effort to ensure similarity or account for existing differences at baseline	No control group
Participant characteristics	Full details about age, gender, ID diagnoses and any other mental or developmental issues, no. of	Partial demographic details e.g. age and gender only	Unclear participant characteristics
Quality of outcome measures	Outcome measures appropriate for use with target population, address the research question, with validation scores	Appropriate outcome measures but not all validation scores documented, or not for specific use with ID	Outcome measures not validated and do not address research aims
Blind treatment assessment	Double blind - participant and facilitator	Single blind - participant or facilitator	No blinding
Missing data	Frequency documented, methods of management stated clearly e.g. reasons for missing post	Missing data documented but management unclear	Unclear or not reported
Treatment effect	Effects are clearly reported with sufficient evidence to support them, i.e. rigorous statistical analysis	Effects are clearly reported but lack sufficient evidence e.g. borderline significance, unclear, or based on	Effects are unclear or unreported
Quality of program facilitator	Details of who the facilitators were, and level of training	Details about facilitator but training not reported	Unclear or not reported
Value of research	All outcomes were considered and can be reliably applied to other relevant populations based on the quality of this research	Alternative outcomes were considered but cannot be generalised or applied to other populations based on this research alone, e.g. small sample size, mothers only, recruitment limitations	Not all outcomes were considered/there is insufficient evidence to apply to other populations
Follow-up	Clear follow-up data > 6 months post intervention completion	Follow-up data < 6 months post intervention completion	No follow-up data/follow-up unclear

Appendix 4: Letter of Access

NHS Ayrshire & Arran

Research &Development Office
58 Lister Street
Crosshouse Hospital
Kilmarnock
KA2 0BB

Miss Lindsey Eunson
Trainee Clinical Psychologist
NHS Lanarkshire
62 Malleny Avenue
Balerno
EH14 7EL

Date 28 January 2019
Your Ref
Our Ref AG/KLB/NM R&D 2018AA056

Enquiries to [redacted]
Extension
Direct line
Fax
Email

Dear Miss Eunson

Letter of access for research: ***Behavioural Family Therapy to support adults with intellectual disabilities: primary caregivers' perspectives***

As an existing NHS employee you do not require an additional honorary research contract with this NHS organisation. We are satisfied that the research activities that you will undertake in this NHS organisation are commensurate with the activities you undertake for your employer. Your employer is fully responsible for ensuring such checks as are necessary have been carried out. Your employer has confirmed in writing to this NHS organisation that the necessary pre-engagement check are in place in accordance with the role you plan to carry out in this organisation.

This letter confirms your right of access to conduct research through NHS Ayrshire and Arran for the purpose and on the terms and conditions set out below. This right of access commences on **28 January 2019** and ends on **1 October 2019** unless terminated earlier in accordance with the clauses below.

You have a right of access to conduct such research as confirmed in writing in the letter of permission for research from this NHS organisation. Please note that you cannot start the research until the Principal Investigator for the research project has received a letter from us giving permission to conduct the project.

You are considered to be a legal visitor to NHS Ayrshire and Arran premises. You are not entitled to any form of payment or access to other benefits provided by this organisation to employees and this letter does not give rise to any other relationship between you and this NHS organisation, in particular that of an employee.

www.nhsaaa.net

INVESTING IN VOLUNTEERS

Appendix 5: Letter of Ethical Approval

WoSRES
West of Scotland Research Ethics Service

NHS
Greater Glasgow and Clyde

Miss Lindsey Eunson
62 Malleny Avenue
Balerno
Edinburgh
EH147EL

West of Scotland REC 3
Research Ethics
Clinical Research and Development
West Glasgow Ambulatory Care Hospital
Dalnair Street
Glasgow
G3 8SJ
(Formerly Yorkhill Childrens Hospital)

Date 08 February 2018
Direct line 0141 232 1807
E-mail WoSREC3@ggc.scot.nhs.uk

Dear Miss Eunson

Study title: **Behavioural Family Therapy to support adults with intellectual disabilities: primary caregivers' perspectives**
REC reference: **17/WS/0258**
IRAS project ID: **234867**

Thank you for your letter (e-mail) of 05 February 2018. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 31 January 2018

Documents received

The documents received were as follows:

Document	*Version*	*Date*
GP/consultant information sheets or letters [GP letter]	1.1	05 February 2018

Approved documents

The final list of approved documentation for the study is therefore as follows:

Document	*Version*	*Date*
Covering letter on headed paper [REC 3 Cover letter]	1.1	11 January 2018
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Public Liability Confirmation]		26 July 2017
GP/consultant information sheets or letters [Therapist Recruitment Letter]	V1.0	10 November 2017
GP/consultant information sheets or letters [GP letter]	1.1	05 February 2018
Interview schedules or topic guides for participants [Interview Schedule]	V1.0	10 November 2017
IRAS Application Form [IRAS_Form_21112017]		21 November 2017

Document	*Version*	*Date*
Letters of invitation to participant [Patient Cover Letter]	V1.0	10 November 2017
Other [Professional Indemnity Insurance]		04 August 2017
Other [Employers Liability Certificate]		01 August 2017
Other [Clinical Trial Liability Insurance]		27 July 2017
Participant consent form [Consent Form Tracked]	1.1	11 January 2018
Participant consent form [Consent Form ER tracked]	1.1	11 January 2018
Participant information sheet (PIS) [PIS tracked]	1.1	11 January 2018
Participant information sheet (PIS) [PIS Easy Read tracked]	1.1	11 January 2018
Research protocol or project proposal [Behavioural Family Therapy to support adults with intellectual disabilities: primary caregivers' perspectives]	V1.0	10 November 2017
Response to Additional Conditions Met [No Cover Letter - Checked in to avoid future reminders]		
Summary CV for Chief Investigator (CI) [L Eunson CV - CI and Student]		
Summary CV for supervisor (student research) [K MacMahon CV]		

You should ensure that the sponsor has a copy of the final documentation for the study. It is the sponsor's responsibility to ensure that the documentation is made available to R&D offices at all participating sites.

17/WS/0258	**Please quote this number on all correspondence**

Yours sincerely

[redacted]

Appendix 6: Therapist Recruitment Email

[insert name]
Trainee Clinical Psychologist
xxxxxxxx@sms.ed.ac.uk
[insert date]

Dear Sir/Madam

Re: Recruitment for study: BEHAVIOURAL FAMILY THERAPY TO SUPPORT ADULTS WITH INTELLECTUAL DISABILITIES: PRIMARY CAREGIVERS' PERSECTIVES

I am currently carrying out a research study to explore primary caregivers' experiences and perspectives of Behaviour Family Therapy (BFT) to support people with intellectual disabilities. As BFT for intellectual disabilities is currently being implemented in your Health Board, I would like to ask for your support in recruiting suitable service users to participate.

Information regarding experiences of BFT will be obtained from the primary caregiver of the person with an intellectual disability using your service. Information will be extracted via qualitative interviews taking place in the caregivers' home or wherever they received BFT. Interviews will last approximately one hour.

I have enclosed a copy of the 'Participant Information Sheet' that gives more details about the study. Please take time to read this.

I have also enclosed the interview schedule and a 'Participant Consent Form'. The 'Participant Consent Form' permits me to use your patient's information and collect data from the interview for analysis and potentially publication and presentation of data. Their

confidentiality will be protected at all times, and any patient identifiable information will be removed from any and all forms of publication or presentation.

If you feel able to assist in this project, I would ask that you first consider potential primary caregivers that would be suitable to take part. The inclusion criteria for participants of this study are as follows;

- The adult for whom they are primary caregiver must have an intellectual disability and additional mental health issues and/or behaviours that challenge
- The adult for whom they are primary caregiver must have received BFT as part of their routine clinical treatment
- They must have attended a minimum of 5 BFT sessions with the person in the aforementioned Learning Disability Services, and progressed beyond the engagement phase of treatment
- They must speak fluent English
- They must be aged 16 or over
- They must understand and be able to consent to take part in the study

If you are interested in getting involved, or would like any more information about the study, then please contact me at the details below.

Thank you very much for all your help.

Yours sincerely

[Insert name and contact details]

Participant Information Sheet

Study Title: **BEHAVIOURAL FAMILY THERAPY TO SUPPORT ADULTS WITH INTELLECTUAL DISABILITIES: PRIMARY CAREGIVERS' PERSPECTIVES.**

You are being invited to take part in a research study that is being carried out by Lindsey Eunson, Trainee Clinical Psychologist, NHS Lanarkshire and University of Edinburgh.

Before you decide whether or not to take part, it is important for you to understand why the research is being done and what it will involve. Please take time to read the following information carefully. Talk to others about the study if you wish. Contact us if there is anything that is not clear or if you would like more information. Take time to decide whether or not you wish to take part.

What is the purpose of the study?

This study intends to explore carers' perceptions and experiences of taking part in BFT. It is hoped that this will give us a better understanding of the things people like and dislike about BFT so that we can improve the service.

Why have I been asked to take part?

Your family has received Behavioural Family Therapy as part of the service provided to your family member who was referred to NHS Lanarkshire's or NHS Lothian's Learning Disability Team.

BFT is a type of talking therapy where we work with families.

When people have problems with their thoughts and feelings, this can make things stressful for them and their family. BFT may help people and their families;

- Talk to each other in a helpful way
- Work out problems together as a family
- Understand each other better
- Support each other to do things they want to do.

Caregivers of people with a learning disability who have received BFT are being invited to see if they want to take part in this study.

Do I have to take part?

No, it is up to you to decide whether or not to take part. If you do decide to take part you will be given this information sheet to keep and asked to sign a consent form. If you decide to take part you are still free to withdraw at any time without giving a reason. Deciding not to take part or withdrawing from the study will not affect the healthcare that you or your family member receives, or your legal rights.

What happens next?

If you want to know more about this study, you can speak to the researcher above or the health workers that you or your family member meets with. You can ask questions at any time.

We will give you this information to take away and read. The next time a member of the team visits you, we will ask you if you have any questions about the study. If you are no longer seeing anyone from the Learning Disabilities Service, the head of your Learning Disabilities service will contact you to ask if you have any questions. Or, you can complete and post the 'indication of interest' form at the bottom of this letter using the prepaid envelope enclosed. We will then answer any questions you may have and ask if you want to take part.

You do not have to take part in the study if you don't want to. If you don't want to take part in the study, it will not affect the treatment

that your family member receives. If you don't want to take part in the study, your family member will still see all the people from the health service that they usually see, like doctors, nurses, psychologists and occupational therapists. It will also not make any difference to any healthcare that you are currently receiving. You don't need to tell us why you don't want to take part.

You can stop taking part at any time. If you stop taking part in the study, your family member will still see the professionals from the Learning Disability service. If you stop taking part in the study, your family member will still see all the people from the health service that you usually see, like doctors, nurses, psychologists and occupational therapists. It will also not make any difference to any healthcare that you are currently receiving. You don't need to tell us why you want to stop taking part.

What happens if I agree to take part?

If you agree to take part in the study, we will;

- Ask you to sign a form to say that you are happy to take part in the study
- Pass your name and contact details to the chief investigator so that they can make contact with you
- Ask you for your GP details so that we can inform them of your involvement in the study
- Make a record of how many sessions you took part in
- Arrange a suitable time to meet and ask you questions about how you found BFT. These questions will be asked by the researcher named above, not by the health workers who completed BFT with you. The health workers that did the BFT with you will not know how you answered these questions. This interview will take place in your home, or where you did the BFT. The interview will take approximately one hour
- To make sure the researcher understood your answers properly, you may be contacted after the interview to have a look over what they have written
- The interview will be transcribed and the study will be completed as part of the researcher's University degree. This study may be published in journals that can be accessed by the general public. Your identity will remain anonymous

What are the possible disadvantages and risks of taking part?

There are no direct disadvantages or risks related to taking part. You would however be required to dedicate one hour of your time for the interview to take place. You may also be contacted to review some of the research to see if it accurately represents your thoughts and feelings about BFT. If you are not comfortable for this to take place in your home, a room at a suitable Health Centre may be booked for this purpose. You are under no obligation to take part. Your participation will have no impact on current treatment for you or your family.

What are the possible benefits of taking part?

Sharing your experiences with us will help us to understand what is helpful and unhelpful about BFT. The information you provide may also lead to service changes and improvements so that more people may benefit from this type of intervention.

What will happen to the information that the research team collect?

The research team will keep all the information that you give us in strict confidence. No one outside the research team will have access to any of your information. The information will be kept very safely on a computer database. Staff will comply with legal requirements for storing data at all times.

The information you share is confidential, but this confidentiality has limits. For example, if you mention that someone is at risk of harm, or that a crime has been committed. All NHS staff have a duty of care to take appropriate action if they are concerned about you or someone that you mention during the interview. They will discuss this with you before doing anything.

What happens after the study is finished?

All the information used in this study will be kept private so that only researchers involved in the study can see it. Any information taken for the study, about you and your family, will not have your name next to it, so no one will know it is about you.

The only person that will see your name, apart from your health care workers, will be the researcher named at the top of this sheet and their supervisor. This is just so we can identify your data so we can remove it if you decide that you no longer wish to take part.

When the study is finished the research team will write about what they have learned so that other people know more about your experiences of BFT. We will let you know what we have found when we have finished the study. Nobody will know who took part in the study because there will be no names or addresses written in the findings.

Who has reviewed this study?

This study is sponsored by the University of Edinburgh. All research in the NHS is looked at by an independent group of people called a Research Ethics Committee. A favourable opinion has been obtained from the West of Scotland Research Ethics Service REC3. NHS Management approval has also been obtained.

If you are interested in taking part or have any questions, please discuss with Lindsey Eunson, xxxxxxxx@sms.ed.ac.uk, 077xxxxxxx in the first instance. If you would prefer, you can ask a member of the Learning Disabilities team who will do their best to answer your questions. Finally, you may also complete and post the 'indication of interest' form below using the prepaid envelope provided if that would be preferable.

If you would like to speak to someone independent of the study team, please contact [insert name] on [insert number], or at xxx.xxxx@lanarkshire.scot.nhs.uk.

If you wish to make a complaint about the study, please contact the University of Edinburgh's Research Governance team via email at: resgov@accord.scot

Thank you for taking the time to think about this study.

Indication of Interest

Study Title: **BEHAVIOURAL FAMILY THERAPY TO SUPPORT ADULTS WITH INTELLECTUAL DISABILITIES: PRIMARY CAREGIVERS' PERSPECTIVES.**

Please tick the appropriate box

I am interested in taking part in this study ☐

I would like to know more about the above study ☐

And agree to be contacted at the details below to discuss this further.

Name:……
……..

Address:……
……

……
……..

Postcode:……
……..

Telephone Number:……

Preferred form of contact (delete as appropriate): Letter / Telephone

Signature:……

Date……..

Participant Consent Form

Study Title: Behavioural Family Therapy to support adults with intellectual disabilities: primary caregivers' perspectives.

Participant Study ID:

Researcher:

Lindsey Eunson, Trainee Clinical Psychologist
s1061346@sms.ed.ac.uk
077xxxxxxxx

Please *initial* each box

2. I confirm that I have read and understand the information sheet (V1.2 dated 23.08.2018) for the above study and have had the opportunity to consider the information and ask questions.

3. I understand that my participation is voluntary and I am free to withdraw at any time without giving a reason, without my family member's medical care or my legal rights being affected.

3. I agree to my anonymised data being used in education publications seen by the general public. All identifiable information will be removed or anonymised. Excerpts of my interview may be quoted in this study.

4. I agree to my interview being audio recorded for transcription and analysis purposes.

5. I agree to my GP being notified about my participation in the study.

6. I understand that relevant sections of data collected during the study may be looked at by individuals from the regulatory authorities and from the Sponsor (The University of Edinburgh) or from the/other NHS Board(s) where it is relevant to my taking part in this research. I give permission for those individuals to have access to my records

7. I understand that my name and contact details will be used by the chief Investigator for contact purposes, for the duration of the study only.

8. I agree to take part in the above study.

_____ _____ _____

Name of participant Date Signature

_____ _____ ___ _____

Name of person taking consent Date Signature

Thank you for taking the time to talk about this study

1 copy for patient, 1 copy for researcher's site file

Appendix 9: Interview Schedule with Supplementary Prompts

1. You and you family did BFT, What was it like?
   - What do you remember about BFT?
   - What did you talk about?
   - How did it you

2. What would you tell other people about BFT?
   - If someone you knew was thinking about doing BFT, what do you think they should know?

3. What was it like working with therapists?
   - How was it meeting at….(home or other session location)
   - How was it having 2 therapists there?

4. Have things changed since doing BFT?
   - What do you think is different now?
   - Are things the same now as they were before BFT?
   - Does the family do anything differently since BFT?
   - Why do you think that is?

Appendix 10: Empirical Study Protocol

Behavioural Family Therapy to support adults with intellectual disabilities: primary caregivers' perspectives

Study Protocol V 1.1

11.09.18

Lindsey Eunson

Author

Contacts Page

Chief investigator: Ms Lindsey Eunson

Trainee Clinical Psychologist

University of Edinburgh/NHS Lanarkshire

Email: s1061346@sms.ed.ac.uk

Academic Supervisors: Dr Ken MacMahon

Senior Lecturer in Clinical Psychology and Clinical Psychologist

University of Edinburgh

Email: ken.macmahon@ed.ac.uk

Field supervisor: Ms Gillian Anderson

Consultant Clinical Psychologist

NHS Lanarkshire

Email: gillian.anderson@lanarkshire.scot.nhs.uk

Contents

List of abbreviations

BFT	Behavioural Family Therapy
ID	Intellectual Disability
IPA	Interpretive Phenomenological Analysis

Study synopsis

Title of study	Behavioural Family Therapy to support adults with intellectual disabilities: primary care givers' perspectives
Study centres	NHS Lanarkshire; NHS Lothian; NHS Fife; NHS Forth Valley; NHS Ayrshire & Arran University of Edinburgh, School of Health and Social Science.
Duration of study	18 months.
Study end point	May 2019.
Objectives	The primary research objective is to explore primary caregivers' perspective and experiences of Behavioural Family Therapy for adults with intellectual disabilities and additional mental health problems and/or behaviours that challenge.
Methodology	Stratified sampling, semi-structured qualitative interviews.
Sample size	There is no specified minimum sample size required for IPA. However research indicates that between 3 and 12 participants are needed to reach an adequate saturation point. It is intended that 12 participants will be recruited, with the expectation that there will be attrition to a final sample size of approximately 10.
Inclusion criteria	• Primary caregivers for an individual with an intellectual disability and additional mental health problems and/or challenging behaviour who have accessed the learning disability service in one of five health board areas (NHS Lanarkshire,NHS Lothian, NHS Fife, NHS Forth Valley, NHS Ayreshire & Arran) AND • Primary caregivers who have attended a minimum of 5 BFT sessions, and progressed beyond the engagement phase of intervention as part of the service user's routine clinical treatment. They may still be engaged in intervention at the point on interview. AND AND • Primary caregivers who have consented to take part in the study AND • Primary caregivers who speak fluent English AND

	• Primary caregivers aged 16 or over.
Exclusion criteria	• Participants who are deemed unsuitable to engage fully in the semi-structured interview process by clinicians (e.g. due to mental or physical health difficulties, problematic behaviours, or intellectual disability).
Statistical analysis	IPA.

Introduction

Research suggests that families who support people with mental health problems are more likely to experience stress. Stress can lead to poorer outcomes, including further mental health deterioration, not just for the individual already suffering from mental health difficulties, but for other family members as well. Behavioural family therapy (BFT) is a form of psychological treatment that has been shown to improve problem solving skills and reduce stress within families. It is commonly used for families experiencing schizophrenia and psychosis, and evidence shows that it is successful in promoting recovery and in reducing rates of relapse. Given these successes, national guidelines now recommend that BFT should be offered to everyone with these disorders. It can however, be used to help families who are experiencing a range of mental health problems such as dementia, depression, anxiety and eating disorders.

Evidence suggests that people caring for someone with an intellectual disability also experience higher levels of stress. Historically, many people with intellectual disabilities were sent to live in long-stay hospitals. However, many of these hospitals were closed following a programme implemented by the Scottish government in the year 2000. Although many regard hospital closures as a step forward, it means that many parents are now expected to care for their adult children in their family home.

People with intellectual disabilities are also at a greater risk of developing mental health problems. There is therefore an increased likelihood that family members caring for individuals with intellectual disabilities will experience high levels of stress and consequential mental health issues. As a result, there has been increasing interest in how BFT may be applied to families caring for individuals who have an intellectual disability and additional mental health difficulties.

Evaluation into how effective BFT is when used in this setting has mainly been completed by services collecting feedback as part of their routine practice. However, there has been one case study published, which detailed a reduction in caregiver strain in someone caring for a person with an intellectual disability, after completing BFT.

Where appropriate, people with intellectual disabilities and their families are currently offered BFT as part of their usual treatment in NHS Fife, NHS Ayrshire & Arran, NHS Forth Valley, NHS Lanarkshire and NHS Lothian. This study aims to explore family experiences of this specific treatment, which may help to shed light on what aspects they found helpful or unhelpful. A previous feasibility study indicates that a sufficient number of caregivers who have received BFT in these health boards may be willing to assist in completing this research.

Despite there being limited evidence for BFT used for people with intellectual disabilities and their families, the research that does exist suggests that it will be a potentially effective treatment. A better understanding of families' perspectives may help to improve the delivery of BFT for other future service users. It may also help to enhance, and ultimately improve the satisfaction and wellbeing of individuals with intellectual disabilities and their families.

Aims

Primary

- To explore primary caregivers' perspectives and experiences of BFT for adults with IDs and additional mental health problems and/or behaviours that challenge.

As this study is exploratory in nature, it is important to avoid any predetermined expectations or hypotheses from impacting the analysis.

Method of investigation

Participants

All participants will be the primary caregiver for the person with an intellectual disability with an additional mental health issue and/or behaviours that challenge. These individuals will be identified by the family and relevant clinician. Participants will be recruited from the NHS Learning Disability Service caseload across NHS Lothian, NHS Lanarkshire, NHS Fife, NHS Ayrshire & Arran, and NHS Forth Valley.

Inclusion criteria

- All participants must meet the following criteria:
- The adult for whom they are primary caregiver must have an intellectual disability and additional mental health issues and/or behaviours that challenge
- The adult for whom they are primary caregiver must have received BFT as part of their routine clinical treatment
- They must have attended a minimum of 5 BFT sessions with the person in the aforementioned Learning Disability Services, and progressed beyond the engagement phase of treatment. Participants still engaged in BFT are also eligible.
- They must speak fluent English
- They must be aged 16 or over
- They must understand be able to consent to take part in the study

Exclusion criteria

- Participants who are deemed unsuitable by relevant clinicians to engage fully in the semi-structures interview process (e.g. as a result of mental or physical health issues, problematic behaviours, or intellectual disability)

Procedure

Step 1 - Awareness: As agreement from therapist to take part in this study has already been obtained, consent forms will not be required from therapist. Therapists delivering BFT from NHS Forth Valley, NHS Fife, NHS Ayrshire & Arran, NHS Lanarkshire and NHS Lothian will however be asked initially to consider potential candidates for the study. They will be given a copy of the patient information sheet detailing confidentiality, the study aims, what the study would involve, and what will happen with the data following completion of the study. This sheet will explicitly highlight that participation or not will in no way affect their routine treatment, and that they are free to withdraw from the study at any time without giving any reason, with no consequence. Therapists may then mention the possibility of voluntary participation in the study to families that will meet the criteria at the time of recruitment.

Step 2 – Recruitment: The database of clients and families who have received BFT as part of routine clinical practice in NHS Lothian, NHS Fife, NHS Forth Valley, NHS Ayrshire & Arran, and NHS Lanarkshire will be accessed by the head of their respective services. Those who completed at least 5 sessions, and progressed beyond the engagement phase will be identified. The primary caregiver for that service user will then be identified by clinicians and confirmed by the

family at the initial contact. A list of primary caregivers who meet all of the inclusion criteria will be compiled. To increase the homogeneity of the sample, recruitment will progress systematically, starting with those who have completed all BFT sessions to those who have attended a minimum of 5, beyond engagement. The length of time since completion will also be taken into consideration at this stage, with those completing most recently being invited to take part first. Those who are still engage in BFT will still be considered, as long as they meet all other criterion.

Primary caregivers from the compiled list will be contacted by letter to ask whether they would be willing to take part in a semi-structured interview concerning their experiences of BFT. The aforementioned information sheet will be included in the letter. An easy read version is available for those who may require it as identified by the relevant clinician. A reply-paid slip will be enclosed for candidates to send back if they are interested in finding out more about the study. The letter will also state that they may return this to a member of the Learning Disability Team, or that they may verbally indicate interest to them. A member of the participants' family members' direct care team will be the person to make initial contact about the study.

If the potential participant is still in contact with the Learning Disability Service, clinicians will discuss their participation at their session following being given the study information. Otherwise, candidates will be given approximately 2 weeks before further contact is made to discuss participation. The researcher will then contact the potential participant by telephone (or in the manner indicated in their reply-slip interest form) to discuss the study and clarify any aspects of participation. Questions will again be welcomed at this stage to ensure that participants are aware of all steps involved in the study process should they wish to proceed.

Step 3 – Informed consent: Primary caregivers who expressed continued interest in taking part in the study will be visited at home, or will come to specified room in a local health care centre to meet with the researcher. Information about the purpose and nature of the study will be reiterated, and questions will once again be welcomed. Should participants choose to continue to take part; a consent form will be provided and completed. An easy read consent form has been developed for those who require it. Consent for the potential use of illustrative interview quotations will be included in this form.

Step 4 – Data collection: Following acquisition of consent, a semi-structured interview will take place. The interview is designed to gather qualitative information about BFT and will consist of open ended questions. The questions may guide participants towards certain areas of interest regarding their experience, but should not encourage concrete responses such as 'good' or 'bad'. A series of predetermined prompts will be used if necessary to encourage elaboration on shorter responses. The first interview will be completed as a pilot interview to explore the suitability of the interview design and questions. If there are no further changes to the interview schedule then this interview will be used in the final data for the study. If changes are required then details of these will be submitted to ethics for approval and the data will be discarded.

Step 5 – Data management: Data will be managed in line with the Data Protection Act (1998), NHS Code of Practice on Protecting Patient Confidentiality (2002), NHS Lanarkshire Information Governance Policies and The University of Edinburgh Data Management Policy.

To maintain confidentiality and anonymity, each consenting participant will be allocated a participant ID. This will be used to represent that individual on all stored data.

Interviews will be recorded using an encrypted NHS device. Recordings will be transferred to an encrypted NHS laptop then deleted from the device following interviews. Recording devices will be stored in locked cabinets on NHS Lanarkshire premises when not in use. All recordings and transcriptions containing identifiable information will be password protected and stored on NHS secure drives. No identifiable information will be held at University sites. Only the researcher and their clinical supervisor will have access to identifiable information. Consent forms will be stored in a locked cabinet at an NHS Lanarkshire secure site. These may only be accessed by the chief investigator and will be stored separately from all other data.

Any notes taking during the interview will be documented to build a picture of the interview environment. When visiting participant homes or health centres, notes, consent forms, and recording devices will be transported using a locked briefcase. Briefcase will be stored in the boot of a locked care and taken directly to and from the specified site.

Confidentiality

Participation in this study is confidential. All identifiable information will be protected and stored only on the central database access solely by the researcher. Recorded data will be anonymised and/or password protected. Raw materials will be deleted 3 years after completion of the study and write up. Responsibility for data deletion will be delegated from the chief investigator to their supervisor at the end of their employment contract. Information regarding confidentiality will be included in the patient information sheet and elaborated on at the initial meeting, before consent is obtained.

Disclosures

Limitations of confidentiality will be highlighted prior to requesting consent. This relates specifically to the Health Care professionals duty of care to take appropriate action should any participant disclose any risk to themselves or someone else, or any criminal activity. Should this type of disclosure occur, participants will be reminded of the limits of confidentiality and that appropriate action will be taken accordingly.

Data analysis

IPA will be utilised to analyse the data obtained from the semi-structured interviews, designed to gather detailed interpretations of each participants' experiences of BFT. IPA was chosen because of its suitability to the exploratory nature of this study. The analysis process will follow that outlined by Smith & Osborn (2007).

The main focus of IPA is to capture the meaning that participants assign to a given experience, in this case their experience of BFT. Thorough interpretation of the data is integral in ensuring that meaning is ascertained accurately. The first step in achieving this is reached through verbatim transcription of each interview. This helps the researcher to fully immerse themselves in the data. This transcript is then read multiple times, whilst simultaneously annotating areas of interest in the left-hand margin of the document e.g. 'appears frustrated' or 'pleasant nostalgia apparent'. Any and all thoughts or interpretations are documented at this stage to avoid any bias, and to allow for future considerations.

Following documentation of initial ideas, the transcript is then read again. Emerging themes are considered at this stage, and reported in the right hand margin of the document e.g. ‘sense of supportiveness’ or ‘emotionally challenging’.

Initial comments from the margins are then consolidated into phrases to represent the overall vibe or tone of the original participant response. More complex, theoretical connections are then deliberated upon, that considers the meaning that participant attached to their comments about BFT. The original transcription is referred to throughout to avoid any outlandish or inaccurate interpretations of responses from being abstracted. All elements of the interview should be addressed equally, avoiding any bias focus of attention of particular sections.

Emerging themes are then listed chronologically based on the interview transcription and connections between them are investigated. Each theme is considered in relation to the transcription as a whole to make sure excerpts are not taken out of context.

A table of themes will then be created for each interview. Clusters of similar themes will be collated to form superordinate themes. Each theme will have a superordinate theme and a supporting excerpt from the original verbatim transcription attached. Some initial themes may be discarded at this point if they seem irrelevant, inappropriate or very infrequent.

This process is repeated from scratch for each participant. It is of the utmost importance that knowledge of previous themes does not contaminate the researchers’ interpretation of future data. To help avoid this from occurring, a second and third researcher will be recruited to assist with a proportion of the analysis. This also helps to

prevent any bias and ensure that an accurate representation of what the primary caregiver said is obtained. Interpretations will also be shared with corresponding caregivers to gain feedback on how accurately their feel the investigator has captured their sentiment.

Risk management

Sense of Obligation

There is a risk of potential participants feeling obliged to take part in the study. Participant Information Sheets will help to reduce this risk, as potential participants will be made aware that there is no obligation to take part, and that withdrawal is acceptable at any time. The information sheet will also highlight that there will be no consequence of refusal or withdrawal, particularly in relation to their routine care. The purpose of the study will be made explicit. The fact that the interviewer was not directly involved in the intervention being discussed should also help to alleviate feelings of responsibility to take part and concerns about offering any potentially negative views on their experience.

Inconvenience

This study hopes to explore experiences of the primary caregivers of an adult with intellectual disability with additional mental health issues and/or behaviours that challenge. Given that research indicates higher stress rates in families caring for someone with an intellectual disability and with mental health issues; there is a risk that asking them to take time out of their duties as a caregiver to participate will increase stress within the family. Caregivers will be made aware that their participation is entirely voluntary and may be withdrawn at any time with no reason necessary.

Misremembering

As we are asking caregivers to recall an intervention that may have many months prior to interview, there is a risk that the information provided will not be as detailed or accurate. This risk will be minimised by ensuring that there are no leading questions on the interview schedule. Consideration of how recently intervention was concluded will be given during the selection process.

Sampling Bias

As eligible participants will be indicated by the head of service, there is a risk that those who are likely to respond positively will be preferred over those who may have indicated a negative treatment experience. Systematic recruitment will help to reduce this risk and discussion around this will precede the selection process. Similarly, eligible participants who have had positive experiences of treatment may be more willing to take part, which may affect the validity of the study and spread of documented experience. Those with negative experiences may be less willing to share these due to fear of the impact this may have on their family member's treatment or how it may reflect back on them. However, the opposite may also be true in that those with negative experiences of BFT may wish to make it known to improve services for others. Attempts will be made by the researcher to welcome all feedback, and to reiterate the inconsequential nature of the study in relation to provided treatment.

Risk to Researcher

Some interviews may take place at the family home, with someone the researcher has never met before. This poses a potential risk to the researcher. Homes will have been visited by other members of the Learning Disabilities team and therefore a full risk assessment will have been completed for that home. The researcher will complete a new updated risk assessment for each participant and will contact clinicians with experience of working with them, to obtain a full understanding of potential risks and how to address them. They will also make sure that another member of staff knows when they are to visit the home, and will arrange a call after leaving where appropriate.

Data protection and confidentiality

There is a risk that participants will be concerned that their information may be inappropriately accessed and personal information may be used if they take part in the study. The Information Sheets will make the arrangements regarding confidentiality explicit. The data being collated will not include any identifiable information and will only be stored in the central database, accessible only to the researcher. Recorded data will be anonymised or password protected. Raw materials will be deleted 3 years after completion of the project and write up.

There is also a risk that participants may divulge adult or child protection issues. All participants will be informed that appropriate protocols and breaches of confidentiality will be followed should these issues arise. It will be made clear that if any concern is raised regarding the safety of a child or adult mentioned throughout the interview process, the relevant services will be informed.

Dissemination

The research will be written up in part fulfilment of the Doctorate in Clinical Psychology as well as being submitted to a peer-reviewed journal. All consenting participants will be informed of the outcome of the study; however, no identifiable information will be shared amongst participants and all findings will be reported anonymously. The results will be summarised in a format appropriate to individuals with an intellectual disability and disseminated throughout each Health Board area involved. The study may also be disseminated at relevant conferences and study days.

Anticipated benefits of the study

As there is very limited evidence to support the use of BFT to support adults with intellectual disabilities, this project may help to build upon the evidence base. As many Scottish Health Boards have now integrated BFT into their standard treatment packages for people with IDs, it would be beneficial to understand more about service users' and their families' attitudes and perspectives on it. Depending on reported experiences, this may help to identify appropriate adjustments necessary to improve service delivery and outcomes and could lead to improved service user satisfaction, and perhaps staff satisfaction as a result.

The intimation of staff currently delivering BFT as detailed elsewhere (Hutchison et al, 2017) is one of optimism, albeit with some frustration. Better understanding the perspectives of service users may reduce frustrations and lead to better communication, understanding and improvement in the general delivery of BFT. The limited existing evidence has promising outcomes, further research may lead to wider spread changes in standard national guidelines for this population i.e. NICE and SIGN guidelines.

Study timetable

Time Period	Process
September 2017	Consult BFT special interest groups and supervision groups for feedback on proposal. Submit ethics application
November 2017	Begin systematic review process
January 2017	Begin recruitment process Conduct pilot interview. Make amendments, resubmit

	ethics if necessary
February 2017 – May 2018	Resume and complete data collection
February 2017 – November 2018	Transcriptions and Data analysis
May 2018 – March 2019	Systematic review and write up
March 2019	First draft
May 2019	Submit final version

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